Original Article

Comparison of congenital and acquired cholesteatomas in pediatric patients

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ABSTRACT

Objectives: This study aims to present clinical data and surgical results of patients with congenital cholesteatoma (CC) and acquired cholesteatoma (AC).

Patients and Methods: Pediatric cholesteatoma patients who underwent tympanomastoid surgery between January 2008 and June 2015 were evaluated retrospectively. Demographic data, clinical symptoms, surgical and post-surgical findings were recorded. Mastoid development was evaluated with preoperative temporal bone computed tomography. Areas with cholesteatoma were mapped intraoperatively and cholesteatoma was staged. Intraoperative stapes superstructure deformation was assessed. Postoperative hearing results were compared according to air bone gap (ABG) values. Statistical analysis was made by Mann-Whitney U and Kruskal Wallis test.

Results: We analyzed 60 patients (9 CC and 51 AC) under 16 years of age and followed at least six months. In the CC and AC groups, mean age was 6.1 and 10.4 years, and mean follow-up was 28 and 32 months, respectively. While CC group patients were generally asymptomatic, AC group patients were diagnosed with complaints of otorrhea, otalgia and hearing loss. Mastoid development was better in CC group patients ($p<0.001$). Intraoperatively, the AC mostly covered two or more regions while CC was mostly in one region. There was more deformity of stapes superstructure in AC group patients ($p=0.019$). Recurrence rates were similar for CC and AC groups and for different types of surgeries ($p=0.128$). Functional postoperative hearing (ABG ≤10dB) was 44.4% in the CC group and 25.4% in the AC group.

Conclusion: The AC is more common than the CC, mastoid development is worse and ossicles are affected more. There are no significant differences in recurrence rates of applied surgeries. However, postoperative hearing results are better in patients who external ear canal preservation, which has an advantage of not causing more mastoidectomy cavity problems. With the main condition of completely removing pathology, these techniques can easily be preferred in cholesteatoma surgery.

Keywords: Acquired cholesteatoma; cholesteatoma surgery; congenital cholesteatoma; pediatric cholesteatoma; tympanomastoidectomy.

Cholesteatoma in children is more aggressive due to anatomic and physiologic differences, and is harder to eradicate.[1,2] There are also many differences between the types of childhood cholesteatoma, congenital (CC) and acquired cholesteatoma (AC). These include differences in pathogenesis, location, spreading pattern and detected age. The pathogenesis of CC is not clear but may be related to embryonic epithelial remnants, while AC develops from

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ventilation deficiency of the middle ear. Clinical presentations of CC and AC are as different as their pathogenesis the former are asymptomatic, while the latter may complain of otorrhea. Therefore, diagnosis of AC is easier than diagnosis of CC. Moreover, while CC is generally limited, AC is extensively spread. The treatment for both CC and AC is surgery,[3] and different surgical approaches are needed because of differences between CC and AC.

It is debatable whether canal wall-up tympanoplasty (CWUT) or canal wall-down tympanoplasty (CWDT) is the best surgical approach to cholesteatoma in children.[4-8] Both procedures have their advantages and disadvantages. With CWUT, the child's ear becomes waterproof and s/he can conveniently engage in water sports like swimming. Hearing is quite satisfactory because the normal anatomic structure is preserved. On the other hand, recurrence rates are higher. The unaesthetic appearance of CWDT might cause psychological problems and certain cavity issues, but disease control is better.[4,7,9]

We aim to examine preoperative, radiologic and surgical differences between CC and AC, surgical procedures in practice, and evaluate recurrence and postoperative hearing results among children with CC and AC in our institution.

PATIENTS AND METHODS

Records of children who underwent tympanomastoid surgery for cholesteatoma in İzmir Bozyaka Training and Research Hospital Otorhinolaryngology clinic between January 2008 and June 2015 were examined retrospectively. Hospital documents were reviewed for age and sex, symptoms, audiometry values, surgical methods, intraoperative cholesteatoma spread, ossicle findings and post-surgery results. Temporal bone computed tomography (TBCT) images were evaluated for mastoid cell development. The study protocol was approved by the Izmir Bozyaka Training and Research Hospital Ethics Committee. The study was conducted in accordance with the principles of the Declaration of Helsinki.

Patients who were diagnosed with pediatric congenital cholesteatoma (CC) and pediatric acquired cholesteatoma (AC), below 16 years old and postoperatively monitored for at least six months were included in the study. Congenital cholesteatoma diagnosis criteria were defined as a white mass that could be seen behind an intact tympanic membrane, no ear discharge or perforation, and no otologic surgery history. A history of otitis media was not a criterion for exclusion.[10] Acquired cholesteatoma patients were classified as retraction pocket cholesteatoma and non-retraction pocket cholesteatoma (cholesteatoma secondary to a tympanic membrane perforation or following trauma or iatrogenic causes).[11]

Surgical techniques carried out for pediatric cholesteatoma in our clinic were CWUT, canal wall-reconstruction tympanoplasty (CWRT) and CWDT. All techniques were executed via post-auricular approach. If the cholesteatoma was small, limited to the middle ear or could be cleared out while maintaining the posterior canal wall, CWUT was performed. If it could not be cleared out this way, the cholesteatoma sac would be followed through the epitympanic recess. If the cholesteatoma lateral border did not exceed the posterior border of the lateral semicircular canal (LSSC), the cholesteatoma would be excised by partially taking out the posterior canal wall, the wall defect repaired with tragal cartilage and perichondrium, and CWRT performed. But if cholesteatoma passed the LSSC posterior border, the facial ridge would be pulled down for proper field of vision, mastoidectomy completed, and CWDT performed with meatoplasty. In all techniques, after the cholesteatoma was cleaned, the middle ear was reconstructed with tympanoplasty and/or ossiculoplasty. Tympanoplasty was done with tragal or conchal cartilage perichondrium. For ossiculoplasty, if the incus was lytic, autologous or homologous incus interposition was performed; if the stapes was lytic, a titanium total ossicular replacement prostheses (TORP-Medtronic Xomed, Jacksonville, FL, USA) was placed between the oval window and tympanic membrane. However, in patients with unhealthy, edematous and pale middle ear mucosa,[12] ossiculoplasty was done in a second stage surgery after six months. All surgeries for patients with pediatric cholesteatoma were executed by the same surgeon.

As a routine in our clinic, when a patient was hospitalized, preoperative TBCT and audiometry tests were performed. Postoperative audiometry was done in the 1st, 6th and 12th months and repeated annually. Using the Japanese Otological Society's (JOS) staging system, these data were evaluated with preoperative TBCT.[13] Intraoperative cholesteatoma expansion was determined by utilizing European Academy of Otology and Neurotology (EAONO)/JOS 2016 classification and staging of middle ear cholesteatoma.[11] Moreover, with the help of JOS 2015 criteria, it was segregated into stages according to the intraoperative condition of the stapes.[13] Postoperative hearing thresholds were analyzed according to the Committee on Hearing and Equilibrium of the American Academy of Otolaryngology Head and Neck Surgery (AAO-HNS) 1995: by accounting for air bone gap at four frequencies (0.5, 1, 2 and 3 kHz).[14] For postoperative hearing analysis, 6th and 12th month audiometry values were taken into account.

Patients were postoperatively followed-up every week in the 1st month, once every two weeks for the next two months, once a month until the 6th month and at the 12th month and annually afterwards. Additional information was acquired from their documents, including follow-up periods, postoperative otorrhea, condition of the cavity, recurrence or whether a second surgery was needed for any reason.

Statistical analysis

Mann-Whitney U and Kruskal Wallis tests were performed by using SPSS version 16.0 in order to evaluate the data (SPSS Inc., Chicago, IL, USA). Significance value was taken as $p<0.05$.

RESULTS

A total of 60 pediatric patients were included in this study. Nine (15%) had CC and 51 (85%) had AC. Of the AC patients, 37 (72.5%) had retraction pocket cholesteatoma while 14 (27.5%) had non-retraction pocket cholesteatoma. There were five (55.5%) male and four (44.4%) female CC, and 30 (58.8%) male and 21 (41.2%) female AC patients. No statistical difference was detected according to gender in both groups (Mann-Whitney U test, $p=0.85$). Mean ages of CC and AC patients were 6.1 and 10.4 respectively. Evaluating both groups according to age, the CC patients were younger and the difference was statistically significant (Mann-Whitney U test, $p=0.012$). The CC and AC patients applied to our hospital for different reasons. Most of the nine CC patients were asymptomatic, with seven (4 referred from other hospitals) diagnosed on routine examination with intact tympanic membrane with a whitish mass behind, and two diagnosed with hearing loss. On the other hand, most of the AC patients presented with symptoms like otorrhea, otalgia, and hearing loss (Table 1). Mean postoperative minimum follow-up periods in CC and AC patients were 13 and 11 months, maximum follow-up periods in CC and AC patients were 49 and 56 months, and mean follow-up periods in CC and AC patients were 28 and 32 months, respectively.

According to CT classification of mastoid cell (MC) development in the CC group, no patient was MC0, two were MC1, three were MC2 and four were MC3. In the AC group, 18 patients were MC0, 24 were MC1, seven were MC2 and two

Table 1. Congenital cholesteatoma and acquired cholesteatoma patients symptoms'

	Congenital		Acquired	
Symptoms	n	%	n	%
Asymptomatic	7	78	4	8
Otorrhea	-	0	14	27.5
Hearing loss	2	22	32	62.5
Otalgia	-	0	1	2

Table 2. Mastoid cell development was better in congenital cholesteatoma group than in acquired cholesteatoma group

	Congenital		Acquired	
MC grade*	n	%	n	%
MC0	-	0	18	35
MC1	2	22	24	47
MC2	3	33	7	14
MC3	4	45	2	4

* MC0: Almost no cell growth; MC1: Cellular structures only around the mastoid antrum; MC2: Well developed cellular structures; MC3: Cellular structures extending to the peri-labyrinthine area; MC: Mastoid cell.

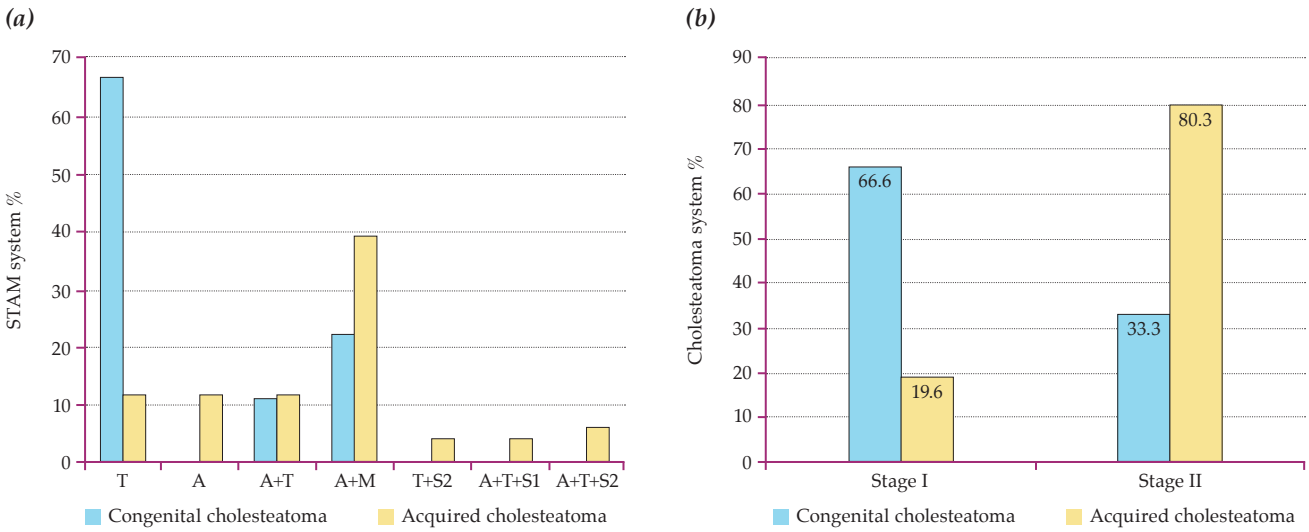

Figure 1. *According to the EAONO/JOS Classification and Staging of Middle Ear Cholesteatoma Criteria,* ***(a)*** *regions of cholesteatoma STAM system (S1: supratubal recess, S2: sinus tympani, tympanic cavity T, attic A and mastoid M)* ***(b)****; staging of cholesteatoma Stage I; Cholesteatoma localized in the primary site, Stage II; Cholesteatoma involving two or more sites.*

were MC3 (Table 2). Mastoid development and pneumatization were markedly better in the CC group (Mann-Whitney U test, p<0.001).

Evaluating cholesteatoma location according to the STAM system (S1: supratubal recess, S2: sinus tympani, T: tympanic cavity, A: attic, M: mastoid), in CC patients six cholesteatomas (66.6%) were T, one (11.1%) was A+T, and two (22.2%) were A+M. In AC patients, six cholesteatomas (11.7%) were T, four (7.8%) were A, 12 (23.5%) were A+T, two (3.9%) were T+S2, two (3.9%) were A+T+S1, three (5.8%) were A+T+S2, two (3.9%) were A+T+S2+M, and 20 (39.2%) were A+M. Using the staging system, in the CC group six patients (66.6%) were Stage I and three (33.3%) were Stage II; in the AC group, 10 patients (19.6%) were Stage I and 41 (80.3%) were Stage II. There were significantly more Stage II patients in the AC group (p<0.001). No patient was classified as Stage III or IV in both CC and AC groups (Figure 1).

According to stapes classification, in the CC group three patients were S0, four were S1, two were S2, and none was S3. Among CC patients, 22.2% did not have a stapes superstructure. In the AC group, 12 patients were S0, nine were S1, 28 were S2, and two were S3. Among AC patients, 58.8% did not have a stapes superstructure (Table 3). The stapes superstructure deformity rate was significantly higher in the AC group (Mann-Whitney U test, p=0.019).

Analyzing surgery types, in the CC group six patients (66.6%) underwent CWUT, two (22.2%) had CWRT and one (11.1%) had CWDT. In five patients only myringoplasty was applied: one removal without tympanic membrane or hearing reconstruction, three incus interpositions, and one TORP. The ossiculoplasty rate was 44.4% and no patient underwent second stage surgery. In the AC group, 38 patients (74.5%) underwent CWDT, seven (13.7%) had CWUT,

Table 3. Intact stapes was better in congenital cholesteatoma group than in acquired cholesteatoma group

	Congenital		Acquired	
S grade*	n	%	n	%
S0	3	33	12	23
S1	4	45	9	18
S2	2	22	28	55
S3	-	0	2	4

* S0: No stapes involvement; S1: The superstructure is surrounded by cholesteatoma and granulation; S2: The superstructure is missing but the footplate remains intact; S3: The footplate is involved and indistinguishable; MC: Mastoid cell.

and six (11.7%) had CWRT. Incus interposition was applied in 30 patients, TORP in 12, and open technique tympanoplasty by supporting round window without ossiculoplasty in 2. Seven patients underwent second stage surgery. Of these seven, a homologous incus was applied in two and TORP in five. The ossiculoplasty rate was 96%. The seven second stage patients have not needed further surgery. In the CC group, only one patient (20%) of those who underwent myringoplasty had recurrence the one who underwent removal without tympanic membrane or hearing reconstruction. The total recurrence rate was 11.1%. In the AC group, four (10.5%) CWDT patients and one (14.2%) CWUT patient had recurrence. The total recurrence rate was 9.8%. There was no significant difference in recurrence rate between CC and AC groups (Mann-Whitney U test, p=0.128).

On evaluating patients according to AAO-HNS criteria of hearing, for postoperative air-bone gap (ABG) values in the CC group, four patients (44.4%) had ≤10 dB, two (22.2%) had 10-20 dB, two (22.2%) had 20-30 dB and one (%11.1) had ≥30 dB. For postoperative ABG values in the AC group, 13 patients (25.4%) had ≤10 dB, 19 (37.2%) had 10-20 dB, 10 (19.6%) had 20-30 dB, and nine (17.6%) had ≥30 dB (Figure 2). Postoperative successful functional results (ABG ≤10 dB) were significantly higher in the CC group (Mann-Whitney U test, p=0.025). There was no significant difference in functional hearing results of patients based on stages and extensiveness of cholesteatoma. However based on stapes classification in the CC group, postoperative functional results were better in S0 compared to S1 and S2 (Kruskal Wallis test, p=0.032), while in the AC group, postoperative functional results were better in S0 compared to S1, S2 and S3 (Kruskal Wallis test, p=0.02).

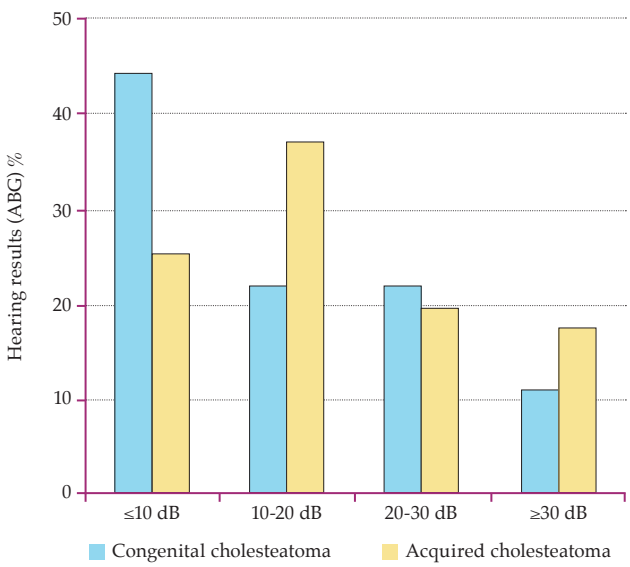

Figure 2. *According to AAO-HNS criteria, achieved functional hearing result was significantly advanced in CC group. ABG: Air-bone gap; AAO-HNS: American Academy of Otolaryngology & Head and Neck Surgery.*

In both CC and AC groups, patients had dry ear almost eight weeks after open cavity surgery and almost six weeks after closed cavity surgery. Only in the AC group did four patients who underwent open cavity surgery have long-lasting postoperative otorrhea, and in these patients, dry ear was achieved in 10-12 weeks with close follow-up and cavity cleaning.

DISCUSSION

Our study examined many characteristics of AC and CC such as diagnosis, treatment period and postoperative findings. Demographically, findings similar to those from the study of Morita et al.[3] were obtained. Both AC and CC group had more male patients. The average age at diagnosis was lower for CC than for AC. While most of the CC patients were asymptomatic (usually presenting with a whitish mass behind the tympanic membrane on routine examination), AC patients were diagnosed with certain clinical symptoms like otorrhea and otalgia. Because pediatric patients cannot verbalize their complaints well, screening otoscopic evaluations ought to be conducted carefully. We speculate that this may reduce the age of diagnosis for CC.

In this study, temporal bone pneumatization and process was better in the CC group than in the AC group. This result supports other similar studies.[3,15] In contrast to the work of Morita et al.,[3] our study showed that intraoperative stapes superstructure deformity was greater in AC patients than in CC patients. Similarly, while stapes superstructure deformity was >40% in the study of McGuire et al.,[7] in our study this rating was almost half in CC and even more in AC. We think that despite the cholesteatoma

being mostly in the tympanic area in CC patients according to the STAM system, the stapes superstructure was not destroyed due to the volume of the cholesteatoma being low. We also think that it might be dependent on cholesteatoma being more extensive in AC patients. The STAM system and cholesteatoma stage data shows that cholesteatoma is more extensive in AC patients than in CC patients. Thus, a more extensive surgery was performed in order to clear out the pathology in AC patients. A CWDT was applied on 11% in the CC group and on 74.5% in the AC group. If we take a look at the ratings of patients who underwent second stage ossiculoplasty, we see that the middle ear and ossicles were affected more in the AC group. In the CC group, the ossiculoplasty rate was 44.4% compared to 96% in the AC group. While none of the patients underwent second stage ossiculoplasty in the CC group, seven patients from the AC group underwent second stage surgery.

When we look at the relationship between extensiveness of cholesteatoma and situation of stapes with postoperative hearing results, we did not detect any apparent effect of extensiveness of cholesteatoma on hearing gain. However, the situation of the stapes affects hearing gain. Patients with unaffected or mildly affected stapes had better postoperative functional hearing.

De Corso et al.[16] suggest second stage surgery on patients due to the high residual disease rates in pediatric cholesteatoma. However, McGuire et al.[7] speculate that patients might not follow the control examination after the first surgery and Roth et al.[6] state that general anesthesia might be traumatic for the child and the parents and thus, do not recommend routine second stage surgery. We also do not routinely implement it; instead we perform second stage surgery on patients who do not have a healthy middle ear mucosa and who will undergo ossicular reconstruction. We did not encounter cholesteatoma recurrence in any of the patients on whom we performed second stage surgery in this study. This supports the idea that second stage surgery is not a routine requirement.

The recurrence rate in CC patients was 11.1%, seen in one patient on whom tympanoplasty was performed. The recurrence rate in AC patients on the other hand, was 9.8%, occurring in 10.5% of the patients that underwent CWDT and in 14.2% of the patients that underwent CWUT. No recurrence was encountered in CWRT patients. Even though the disease is more extensive, recurrence rates are relatively low in AC patients. Because our CC patients count was low, we suppose comparing recurrences will not be appropriate. In addition, having performed more extensive surgeries on AC patients might have reduced recurrence rates. The fact that there is no significant difference between CWDT and CWUT recurrence rates in AC patients and that no recurrence was seen in patients that underwent CWRT shows that with appropriate patient selection, surgeries that preserve the anatomy and provide low recurrence can be performed. The selection of type of surgery depends on several factors such as extent of disease, social status of the patient, hearing level of the patient, and experience of the surgeon.[6] Whatever the method chosen, a successful surgery should achieve total removal of unhealthy tissue and rehabilitation of hearing. It should also prevent recurrence.[6,7]

Cholesteatoma is a disease that has to be followed postoperatively for many years. There is no consensus about the duration of follow-up after cholesteatoma surgery in children. However, long-term monitoring is considered to be critical and findings like postoperative recurrence and hearing results are thought to be changed with respect to follow-up duration.[4,5,17]

Air-bone gap values less than or equal to 10 dB which provide postoperative functional hearing, were 44.4% in CC and 25.4% in AC patients, respectively. Postoperative functional hearing results in the CC group can be associated with both the patients' insignificant preoperative hearing losses and performing surgeries in which the posterior canal wall was preserved in most patients. Moreover, when we examine the hearing results according to the executed surgical procedure, we see that in both groups, functional hearing was provided in greater proportion in surgeries where the ear canal wall was preserved (CWUT and CWRT).

The primary purpose of cholesteatoma treatment is to eradicate pathology. Therefore,

performing CWDT should not be avoided if needed. However, when we review recurrence rates or hearing results, we see that CWRT can be conveniently applied on patients whose pathology could not be eradicated with CWUT in cholesteatoma surgery. Canal wall-reconstruction tympanoplasty not only gives us the advantage of exposure of the CWDT technique, but also provides normal anatomy as in the CWUT technique.[18] Further, it avoids creating the open cavity that could cause psychological and cosmetic problems. Patients can conveniently do various water sports like swimming.

The most important limitation of our study is the small number of CC group patients compared to number of AC group patients. We need further studies that include larger numbers of patients.

Conclusion

In this study, we observe that AC progresses more extensively, mastoid development is worse, and the ossicles are affected more than in CC patients. However, with appropriate surgery selection, a dry ear can be achieved in both. Also, both CWUT and CWRT patients had low recurrence rates and fine hearing results. With appropriate patient selection, CWRT can be implemented as an alternative to CWDT in cholesteatoma surgery.

Declaration of conflicting interests

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