

Efficacy of an Exclusive Transcanal Endoscopic Ear Surgery for Congenital Cholesteatoma



Abdulrahman K Aledan* and **Ebraheem A Alnofal**

Department of Otolaryngology, King Saud Medical city, Riyadh, Saudi Arabia

Submission: September 03, 2019; **Published:** September 25, 2019

***Corresponding author:** Abdulrahman K Aledan, Department of Otolaryngology, King Saud Medical city, Riyadh, Saudi Arabia

Abstract

Background: Congenital cholesteatoma is a type of cholesteatoma; it is a defect developed during embryonic stage and occur by entrapped of the epithelia in the middle ear cleft Endoscopic approach recently was used for the mastoid and middle ear. Development in endoscopy has been performed.

Aim: To investigate the ability to use endoscopic ear surgery exclusively for congenital cholesteatoma.

Methods: Searching process in the Pubmed database was performed to obtain articles related to our subject. Several keywords were used to obtain all possible articles. The included articles were chosen according to specific criteria.

Results: There were nine articles included in this article review as they met the inclusion criteria and 21 articles were excluded. The results of the articles were summarized according to the main titles in the table.

Conclusion: Endoscopic ear surgery is a successful exclusive strategy for the management of pediatric congenital cholesteatoma.

Keywords: CC; CC management; Endoscopy; Pediatrics

Introduction

Cholesteatoma is the growth of abnormal keratinizing squamous epithelium with the combination of cystic lesions, cystic mass with surrounding inflammatory reaction, keratin debris, three-dimensional structure, middle ear tumor, epidermic cyst and form of chronic otitis media [1-10]. Cholesteatoma is classified primarily into acquired and congenital [11]. Acquired cholesteatoma (AC) is divided into primary and secondary types according to the migration of the epithelium into the middle ear and presence of perforation [6,11].

Whereas congenital cholesteatoma (CC) is a defect developed during embryogenesis when the epithelial rest become entrapped in the middle ear cleft [12]. In other word, CC is described by its presence in the middle ear cavity [12]. Clinical presentation of CC is based on the extent and location of the lesion [13]. CC is characterized by white mass medial to the tympanic membrane, abnormal otoscopic examination, conductive hearing loss and ear pain as well as neck pain. However, this pain rarely occurs [13,14]. Otorrhea and otalgia are rare, whereas 50% of patients had a history of previous otitis media episodes [15]. The prevalence of CC is low ranging between 2% to 5% of cholesteatomas [16]. However, the higher

prevalence range among children was estimated to be 4% to 24% of all cholesteatoma's cases [17].

Staging systems were established to enhance and increase the treatment outcomes in CC children patients; there are two staging systems proposed [18-20]. The staging system by Nelson et al. [19] involves two types of CC, type 1 which involves the middle ear with ossicular involvement, whereas type 2 involves ossicular involvement defined as ossicular erosion or removal of ossicles due to disease. The staging system by Potsic et al. [18] involves four stages of CC, stage 1 refers to a single-quadrant disease, whereas stage 2 and 3 involves disease in multiple quadrants and ossicular involvement respectively, whereas the fourth stage of the disease is defined when the disease is any mastoid extension. Surgery is the treatment of CC, and the goal of this treatment is to remove the disease completely with the restoration of hearing, the conventional treatment includes canal wall down or intact canal wall combined with tympanoplasty [21].

Recently, endoscopic approach to the mastoid and middle ear was used by several investigators to reduce the morbidity of a second look operation [22]. Endoscopy became available

in Europe in the late 1970s for evaluation of patients' ears and rarely for diagnostic purpose in operation rooms [23]. The first endoscopically guided surgery was performed in 1987 in France to prevent residual cholesteatoma [24]. The reduction of morbidity and increase in surgical procedure accuracy were achieved using endoscopy [25]. The role of the endoscope in the management of cholesteatoma, especially in pediatric patients has been developed [26-28]. The endoscope was used combined with a microscope resulting in a lower rate of residual disease [28].

Moreover, the endoscope was incorporated as the primary method for visualizing the operative field during surgical dissection of cholesteatoma from the middle ear [29]. Several advantages of endoscopic ear surgery in pediatric patients were reported by Tarabichi, and they included a dynamic and broad view of the middle ear with the ability to "see around corners," improved visualization reduction in the postauricular incision and dissection rates [30]. However, several drawbacks were stated, and they included potential increased initial surgical times and cost, the loss of binocular vision as well as a learning curve to master one-handed endoscopic surgical technique [29].

The present article review was conducted to investigate if the endoscopic ear surgery can be exclusive for CC.

Materials and Methods

This article review is presented to investigate if endoscopic ear surgery can be exclusively used for CC by reviewing previous articles. The search process was performed using the Pubmed database and several keywords to obtain related articles including "Congenital Cholesteatoma, CC management, Endoscope, Endoscopic ear surgery". We obtained 30 articles, which were selected according to specific criteria, these criteria include Congenital Cholesteatoma mainly, exclusive use of endoscopic surgery, follow up after surgery and include recurrence, residual disease, audiology and complications and pediatric patients however any age group can be included, the exclusion criteria included pure use of microscopic ear surgery and only use of endoscopic for assessing microscope ear surgery without comparing. After reviewing abstracts of the articles and according to criteria chosen, only nine articles were included, and the other articles were excluded. The publication years of included articles were between 2018 to 2008.

Results

Table 1: Author and publication year, study design, number of patients and main characteristics, cholesteatoma type/ classification, endoscopic strategy and follow up as well as results and main findings.

Author and Publication Year	Study Design	Number of Patients and Main Characteristics	Cholesteatoma Type/ Classification	Endoscopic Strategy and Follow Up	Results and Main Findings
Park et al. [31]	Retrospective review	25 pediatric patients with CC, whose age range was 17 months to 9 years	*Stage I:13 patients	Transcanal endoscopic ear surgery (TEES) by experienced surgeon	* No surgical complications occurred
			*Stage II:7patients	-Follow up for 24 months	* Recurrence occurred for one case with stage II
			*Stage III:3patients		* Ossicular reconstruction was performed for 2 patients whose stage was II
					*CC limited to middle ear cavity could be removed successfully by TEES in all patients.
Kim et al. [32]	Retrospective case review	12 children with CC involving anterior surface of the malleus and tensor tympani (ASMT), whose age range 2-10 years	*Stage I: 8patients	TEES for 3 patients	*ASMT was well visualized by endoscope assistance in all cases
			*Stage II:2 patients	-Endoscopic assisted surgery for 9 patients	*No hearing deterioration at 3
			*Stage II:2 patients	- Follow up mean duration 15.5 months	months after surgery
					*No residual or recurrent disease during the follow-up period
					* No complications
		*Endoscopic ear surgery should be considered in patients with CC in the ASMT			

Ghadersohi et al. [29]	Retrospective case series	65 pediatric C cases in 38 ears	* Not mentioned	*Endoscope ear surgery (EES) was performed for 34 patients (38 ears)	* Surgical time was longer for acquired cases (226 min Vs 154 min)
		*Acquired C=27 ears (age mean 10.9 years)		-Endoscope was used as the primary visualization tool in 31 ears	*Recurrence in 5 ears, residual disease in 4 ears (3 acquired and 1 congenital)
		*CC= 11 ears (age mean 3.8years)		- Endoscope was used as an adjunct to the microscope in seven ears	* Hearing outcomes were comparable for both cholesteatoma types.
				-TEES for 22 years and 35 cases	* The lowest rates of recurrent and residual disease were seen in EES 3 cases
				-Follow up for 2.6 years (9 months to 4.6 years)	* Low rates in EES 2 and 3 ears, including four recurrences and two ears with residual disease
					* The endoscopes are a viable tool for resection of pediatric cholesteatoma and provide excellent visualization of the middle ear and associated recesses.
Machiono et al. [33]	Retrospective chart and surgical video review of patients with bilateral congenital cholesteatoma (BCC)	6 pediatric patients with BCC with a median age of 4 years (range:2-7 years)	* Not mentioned	Transcanal endoscopic approach (TEA) was performed for 6 ears	* No complications in any patients
				- microscopic post auricular tympanoplasty was performed for 2 ears	* The endoscope allows good surgical control of cholesteatoma removal from hidden recesses.
				-Canal wall up mastoidectomy for 4 ears	* The TEA can have a valuable role in BCC
				- Follow up mean duration 54.5 months	
Cohen et al. [34]	Case series with chart review	55 pediatric patients (56 ears) with median age of 7.7 years and range of 6.7-13 years	*Stage I:7	Endoscopes (EES) were used for inspection in 25 primary resections and for dissection in 39 primary resections.	* Extent of disease at the time of primary resection was similar among groups
		Patients were divided into inspection group (24) and dissection group (32)	*Stage II : 8	-Follow up duration wasn't mentioned, but it wasn't long	* Rate of mastoidectomy significantly decreased from 63% to 33% over the study period with no significance (P = .04) with similar disease extent
			*Stage III: 25		* There was no significant difference in residual disease between those performed EES for inspection and for dissection.
			*Stage IV : 16		* Residual cholesteatoma rates during planned second-look procedures were similar between the study groups.
					* Use of the endoscope led to a lower rate of mastoidectomy
					for cases with similar disease extent.

Hunter et al. [26]	Case series with chart review	66 patients (76 ears) who underwent cholesteatoma (range 4-18 years)	* Not mentioned	Microscopic removal of cholesteatoma: 74 ears	*Though second-look procedures occurred in 15 endoscopic cases and 10 microscopic cases (P = .006)
		removal with either endoscopic or microscopic visualization		-Endoscopic+Microscopic removal: 29 ears	*The rate of residual disease didn't significantly differ in endoscopic and microscopic cases (20.0% and 40.0%) respectively
		with mean age of 10.9 years		-TEES: 8 ears	*only the air-bone gap for TEES demonstrated significant
				-follow up for 18.8 months (range 6.7-48.3)	improvement
					*No complications
					*Both of endoscopic cholesteatoma surgery and traditional microscopic techniques have similar hearing outcomes, rates of recurrence and residual disease, and complications
Kobayashi et al. [35]	Retrospective case review	12 children with CC confined to middle ear, whose age range was 1-6 years	*Stage I :7	Transcanal middle ear surgery using endoscopes	*The one with stage III had residual diseaseduring a second-look procedure
			* Stage II:4	-TEES for 12 patients	*other patients performed one-stage operation and didn't suffer recurrence
			*Stage III:1	-Microscopic surgery for 4 patients during the same period	*early-stage CC can be safely removed using TEES.
				- Follow up mean duration was 23.1 months	
Machironi et al. [36]	retrospective chart review	54 children with median age 9.6 years (range 4-16 years)	*Cholesteatoma staging was as follow; attic (A), tympanic cavity (T) and mastoid (M) sites, with a specific identification of the facial recess and/or sinus tympani extension (S)	TEA was performed for first group	*Recurrence rate was 12.9% (4 ears) for the transcanal endoscopic approach group and 17.2% (5 ears) for the canal wall up microscopic approach.
		Divided in to 2 groups:		-Canal wall up microscopic technique (CWU) was performed for the second group (control)	*Residual disease was present in 26.6%: 19.3% (6 ears) for the transcanal endoscopic approach group and 34.4% (10 ears) for the canal wall up microscopic approach.
		*First group: patients presenting with cholesteatoma involving the tympanic cavity (mesotympanum and epitympanum), attic, retrotympanic and/or protympanic and/or hypotympanic regions, with no antral/ mastoid involvement		-Follow up mean duration was 36 months (range8-88)	*Analysis at 36 months showed a lower recurrence risk for the transcanal endoscopic approach compared with the canal wall up microscopic approach, but with no significance
		*Second group (control): patients with mastoid extension of the pathology			*TEA represents a feasible, minimally invasive and conservative technique for the management of pediatric middle ear cholesteatoma
		-Bilateral pathology was found in 5 cases, giving a total of 59 ears undergoing surgery			

Kojima et al. [37]	Case report	One female patient with 26 years old who suffered hear loss in the right ear	-Not mentioned	308 and 708 rigid endoscopes were used for the operation via the middle cranial fossa.	*The inferior surface of the cochlea and the region around the internal carotid artery could be well visualized by use of endoscopy
				-follow-up 3 years	*petrous cholesteatoma surrounding the cochlea successfully and completely removed with preserving preoperative hearing
					*Endoscope-assisted surgical technique that allowed safe and complete removal of a cholesteatoma extended inferior surface of coclear and around carotid artery in the petrous.

After selecting the articles according to the inclusion criteria, we reviewed the abstracts and the full articles, and we summarized them under specific titles as follow; table 1. According to publication year, there were 2 articles published in 2018 [31,32], 3 published in 2017 [29,33,34], one in 2016 [26], 2 in 2015 [35,36] and 1 in 2008 [37]. There were 6retrospective studies, one retrospective review [31], 2 retrospective case reviews [32,35], one retrospective case series [29], one retrospective chart [33], and one retrospective chart review [36], and 2 case series with chart review [26,34] and one case report [38,39].

The total number of patients in this article review is 296. However, the number of ears was more than a few patients as many patients suffered bilateral disease. The age of all patients didn't exceed 18 years old, except for one study that was case report performed on the female with 26 years old [37]. There were 5 studies that didn't divide patients into groups, one included 25 children with CC and age range 17 months to 9 years [31], one included 12 children with CC with age range 2-10 years [32], one included 6 children with bilateral CC with age range 2-7 years [33] and the last one included 12 children with CC and age range 1-6 years [35].

The other 5 studies, one divided patients into 2 groups according to type of cholesteatoma into AC and CC [29], another one divided patients according to surgical strategy into inspection group and dissection group [34], another study divided patients according to strategy used for surgery into endoscopic and microscopic [26], another study divided patients according to pathology into control group and cholesteatoma group [36], the last study was case report included one female who were suffering petrous cholesteatoma [37]. The type/ classification of cholesteatoma wasn't mentioned in 4 studies [26,29,33,37], the other 5 studies mentioned the classification of cholesteatoma, 3 studies included patients with stage I, II and III [31,32,35], where patients in stage I were more dominant in the 3 studies, one study reported presence of patients with stage IV, but with more presence of patients with stage III [34], one study used the stage of cholesteatoma according to its site [36].

Regarding endoscopic strategy performed, TEES was performed for all patients in one study [31], TEES was performed

for 3 patients in another study [32], for 35 cases [29], TEA was used for 6 years in one study [33] and one group of31 patients [36], TEES was performed for 8 ears [26], and 12 patients [35]. The range of follow up duration in all studies was 6.7 months-7.3 years, only one study didn't mention the exact duration of follow up, but they mentioned that the follow-up duration wasn't long [34]. The main findings and results reported that no complications occurred [26,31,32,33], recurrence occurred in one patient with stage II [31], and residual disease in one patient with stage III[35], recurrence and residual disease reported in4 studies [26,29,34,36], however the lowest recurrence and residual disease rate was found in those performed endoscopy in2 studies [29,36] and they were the same rate as other techniques in2studies [26,34].

The endoscopic was reported to be superior in 6 studies to manage CC successfully in pediatrics [29,31,32,33,35,36], while one study reported its successful role in visualization [29], whereas 2studies showed it wasn't superior and resulted in similar results as conventional strategies [26,34]. Moreover, the case report showed that endoscope assisted surgical techniques to be efficient in removing of petrous cholesteatoma [37].

Discussion

Successful of endoscopy in adults were reported in previous studies. It was reported in one study in 1995, that endoscopically assisted ear-surgery was beneficial in 9 cases and reduced the morbidity. Also, it increased the accuracy of the surgery [38]. On endoscopy evaluation, low rate of residual cholesteatoma, as well as no false negative, was reported in one study in 1997 [39]. Also, by using endoscopy as second look following primary conventional procedure showed a lower rate of residual disease than conventional methods and there was no false negative in patients undergoing endoscopic surgery [40]. The present article review was conducted to evaluate if endoscopic ear surgery can be exclusive for pediatric CC.

We reviewed nine articles; they were either retrospective reviews or case series, only one case report was included. However, this case report was for the adult with petrous cholesteatoma. The number and age of children patients were highly varied between the different studies, whereas the

classification of cholesteatoma was too close among included studies. Several interventions were used for management of CC, either endoscopic alone, or combined with other intervention. Findings of most studies showed that endoscopy was a successful treatment for complete removal of pediatric CC.

Moreover, it was also an excellent visualization method. Only two studies showed that endoscope wasn't superior in the management of pediatric CC [26,34]. One study on pediatric with chronic ear problems was conducted to evaluate the accuracy of the endoscopy, and it was concluded that "open second-look mastoidectomy might be avoided if minimal or no cholesteatoma is found during the endoscopic exploration at the second look" [41]. One previous study reported the effectiveness of endoscopy approach in reducing the rate of residual cholesteatoma and improving the hearing threshold [42].

Conclusion

Endoscopic ear surgery is a useful and successful strategy for pediatric CC management. However early detection is the best way for good outcomes. No, complications for endoscopy were reported and regarding recurrence as well as residual disease, endoscopy was better to decrease the rate of both, only a few reports showed similar rates with other conventional strategies

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DOI: [10.19080/GJO.2019.20.556049](https://doi.org/10.19080/GJO.2019.20.556049)

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