International Journal of Pediatric Otorhinolaryngology 79 (2015) 2316–2320

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Transcanal CO_2 laser-enabled ablation and resection (CLEAR) for intratympanic membrane congenital cholesteatoma

Chang-Ho Lee, Ji Yoon Kim, Young Ju Kim, Chan Kee Yoo, Hyoung-Mi Kim, Jae-Cheul Ahn *

Department of Otorhinolaryngology – Head and Neck Surgery, CHA Bundang Medical Center, CHA University, Seongnam, South Korea

ARTICLE INFO

Article history:
Received 17 August 2015
Received in revised form 23 October 2015
Accepted 26 October 2015
Available online 2 November 2015

Keywords:
Cholesteatoma
Congenital
Lasers
Tympanic membrane
Keratins

ABSTRACT

Objectives: Cholesteatoma in the tympanic membrane is frequently regarded as congenital but there has been no case series review or comparison study with typical pediatric congenital cholesteatoma (CC).
Methods: All pediatric CC cases from 2009 to 2014 were collected, and a total of 10 cases of intratymapnic membrane CC (ICC) out of 429 CC cases were reviewed. They were compared with 14 cases of iatrogenic intratympanic membrane keratin after pediatric CC surgery (IIKC).
Results: ICC constituted 2.3% (10/429) of CCs, and the median age of operation was 24 months, 12 months earlier than that for CC. ICC failed to show male preference which is found in both CC and IIKC. As CC is commonly abutting the medial side of malleus, almost 90% of ICCs were found abutting the umbo of malleus. However, IIKC was usually located at the epithelial trauma site during the CC surgery without malleus abutment. Except in case of spontaneous resolution, the other cases of ICC and IIKC were treated by minimally invasive transcanal CO_2 laser-enabled ablation and resection (CLEAR) alleviating any ossicle vibration trauma, incision or graft harvest. All patients retained normal hearing without complication and recurrence.
Conclusion: ICC might be a rare variant of early detectable pediatric CC estimated from its location close to the ossicle and the surgical findings, but without sex preference. Although ICC shares morphologic similarity with IIKC, their locations of development are different, thereby suggesting different pathogenesis rather than epithelial injury. However, CLEAR surgery can be a good treatment option for intratympanic membrane lesions.

1. Introduction

Congenital cholesteatoma (CC) is incidentally found as a whitish mass medial to the normal tympanic membrane during otoscopic examination, which is diagnosed if the child has (1) no evidence and history of otorrhea or perforation and (2) no prior history of otologic surgery or trauma. The evidence that these lesions are congenital in origin is based on clinical findings; the young age of presentation, the normal appearance of the middle ear mucosa in almost all cases of small lesions, normal mastoid pneumatization documented by computerized tomography, the relatively large number of children with associated minor or major congenital malformations, and the consistent male preference [1].

Intratympanic membrane congenital cholesteatoma (ICC) is regarded as a rare variant of CC in children [2], which is credited in the literature as diagnosis such as; CC in the tympanic membrane [3,4], CC of the tympanic membrane [5], and intramembranous tympanic membrane CC [6]. However, since there are limited case reports, it was unclear whether ICC showed the same clinical evidence as CC mentioned above. Since ICC is located inside the tympanic membrane, it does not readily follow the definition of CC which is found medial to the membrane. Its congenital entity was sometimes denied to be a intratympanic membrane cholesteatoma [7] or tympanic membrane keratoma [8]. ICC is morphologically and positionally similar to iatrogenic intratympanic membrane keratin pearl after CC surgery (IIKC), which is not congenital, and ICC was once explained by Ruedi's theory as arising from inflammatory injuries to the squamous epithelial basal layer [9].

In this study, we aimed to describe our experience of ICC and IIKC in children and to analyze their characteristics and treatment outcomes. Furthermore, in order to find distinct characteristics of ICC and IKC, this study compared ICC, IIKC, and standard CC.

* Corresponding author at: Department of Otorhinolaryngology – Head and Neck Surgery, CHA Bundang Medical Center, CHA University, 59 Yatap, Bundang, Seongnam 463-712, South Korea. Tel.: +82 10 9052 1202; fax: +82 31 780 3449.
E-mail addresses: jcahn@cha.ac.kr, myblueart@naver.com (J.-C. Ahn).

http://dx.doi.org/10.1016/j.ijporl.2015.10.035
0165-5876/

2. Materials and methods

2.1. Study design and population

This study presents consecutive cases of ICC and IIKC in children who underwent operation for CC in a university hospital from January 2009 through December 2014. ICC is defined as a whitish mass on the tympanic membrane without prior history of otorrhea, tympanic perforation or previous otologic procedure [1]. IIKC was defined as a newly developed whitish mass on the tympanic membrane after surgery for CC. This study included a pediatric patient (age < 72 months) with ICC and IIKC. During the study period, there were 429 consecutive cases of pediatric CC. ICC constituted 2.3% (10/429) of the congenital cholesteatoma, which was not significantly different to the 3.3% incidence of IIKC (14/429) after surgical trauma. None of IIKC was developed from ICC.

2.2. Surgical method for ICC and IIKC

The ICC and IIKC were treated with trans-speculum CO_2 laser enabled ablation and resection (CLEAR) with Steri-strip® patch, without any endaural incision (Supplemental Video). All surgery was performed by a single surgeon, C.H.L., who has surgical experience of about 500 cases of congenital cholesteatoma. In order to evaluate the invasion into the middle ear cavity, preoperative high resolution (0.625 mm thickness) temporal bone computed tomography (TBCT) was taken for all patients with ICC.

Before the CLEAR operation, topical anesthesia was performed with a piece of a Gelfoam® (Pfizer, New York, NY) soaked with Xylocaine (AstraZeneca, Seoul, South Korea) for 30 min. After positioning the speculum in the affected ear canal, the tympanic membrane was examined under a microscope OPMI PROergo/S7 (Carl Zeiss, LLC, USA). The outer epidermal layer of the tympanic membrane was vaporized with a CO_2 laser with an Acuspot® 712 micromanipulator (Sharplan, Allendale, USA) in 3W continuous mode. The cholesteatoma pearl was vaporized until sufficient decompression came to plausible evacuation, while preserving the inner fibrous layer of the tympanic membrane. Complete vaporization of the marginal epidermal sprout should be conducted to prevent recurrent intratympanic keratin entrapment. After vaporization, a piece of Steri-strip® was used to completely cover the wound on the tympanic membrane.

After CLEAR operation, children visited the clinic within 2 and 4 weeks to confirm complete healing of the perforation: they were examined with otomicroscopy at 2 weeks and impedance audiometry was obtained at 4 weeks. With confirmation of the healing, Steri-strip® patch was removed.

2.3. Statistics

This study reviewed the medical records of consecutive cases during the study period, analyzed, and summarized retrospectively the results. The size of the lesion was analyzed for demographic and clinical parameters; sex, age at operation, duration until revision surgery, and the affected side. The analyses were performed by Fisher exact test and Mann–Whitney test for categorical parameters and linear regression for continuous parameters. Statistical significance was defined by a two-tailed p-value < 0.05.

3. Results

3.1. Characteristics of ICC and IIKC

A total of 24 children were enrolled in this case series; 10 with ICC and 14 with IIKC. The enrolled 24 children consisted of 18 boys and 6 girls. In ICC cases, there were 5 boys and 5 girls while there were 13 boys and 1 girl in the IIKC cases. The median age at the operation was 24.0 months for ICC cases. The median age of IIKC cases was 11.5 months (range 6–15 months) after the initial surgery, median age of which was 43.0 months. The demographic and clinical findings of the cases were summarized in Table 1 and 2.

The median sizes of the ICC and IIKC were 3.5 mm and 3.0 mm, respectively, (Table 2). The size was independent of sex for both ICC ($p = 0.915$) and IIKC ($p = 0.687$); the median size was 4.0 mm and 3.0 mm for boys and girls in the ICC cases and 3.0 mm for both

Table 1
Patient profiles of intratympanic membrane congenital cholesteatoma (ICC) and iatrogenic intratympanic membrane keratin after congenital cholesteatoma surgery (IIKC).

Patient number	Sex	Age (month)	Diagnosis	Side	Location	Size (mm)	Treatment
1	Male	16	ICC	Right	Umbo	1	None[a]
2	Male	51	ICC	Left	Umbo	2	CLEAR
3	Female	24	ICC	Left	Umbo	2 (triplet)	CLEAR
4	Male	24	ICC	Left	Umbo	4	CLEAR
5	Female	38	ICC	Right	Umbo	2	CLEAR
6	Female	17	ICC	Right	Annulus	3	CLEAR
7	Female	30	ICC	Left	Pars flaccida	4	CLEAR
8	Male	24	ICC	Left	Umbo and PSQ	5	CLEAR
9	Female	13	ICC	Right	Umbo and PSQ	6	CLEAR
10	Male	35	ICC	Right	Umbo, AIQ, and PIQ	6	CLEAR
11	Male	62	IIKC	Right	PSQ	1 (doublet)	CLEAR
12	Male	55	IIKC	Left	Annulus	3	CLEAR
13	Male	40	IIKC	Left	PSQ	1	CLEAR
14	Male	61	IIKC	Left	Annulus	1	CLEAR
15	Male	46	IIKC	Right	Annulus	3	CLEAR
16	Male	37	IIKC	Right	ASQ	4	CLEAR
17	Male	55	IIKC	Left	ASQ	3	CLEAR
18	Male	54	IIKC	Right	ASQ and PSQ	1 (doublet)	CLEAR
19	Female	49	IIKC	Right	PSQ	3	CLEAR
20	Male	55	IIKC	Left	PSQ	3	CLEAR
21	Male	41	IIKC	Left	ASQ	3	CLEAR
22	Male	47	IIKC	Left	Annulus	2 (doublet)	CLEAR
23	Male	60	IIKC	Right	Pars flaccida	3	CLEAR
24	Male	66	IIKC	Right	PSQ	4	CLEAR

[a] Spontaneous rupture and resolution.
AIQ: anteroinferior quadrant; ASQ: anterosuperior quadrant; CLEAR: CO_2 laser enabled ablation and resection; PIQ: posteroinferior quardrant; PSQ: posteriosuperior quadrant.