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Clinical characteristics of pediatric external auditory canal cholesteatoma

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ABSTRACT

Background & objective: External auditory canal cholesteatoma (EACC) is caused by an invasion of squamous tissue into a localized area of periosteitis in the bony canal wall. The clinical characteristics of pediatric EACC are still unknown because of its rare occurrence. To date, only a single paper has reported that pediatric EACC has a less aggressive growth pattern compared to adult EACC. Further studies are required to understand the clinical behavior of EACC, i.e., its aggressiveness. The purpose of this study was to evaluate the clinical characteristics of pediatric EACC.

Materials and methods: The clinical records of all patients diagnosed with EACC in our department from January 1, 2012 to February 29, 2016 were retrospectively reviewed, focusing on the extension of bone erosion, symptoms, and clinical findings.

Results: Seven patients had primary pediatric EACC (age range, 5–17 years). All patients showed unilateral EACC. Otalgia and intermittent otorrhea were common symptoms. Bacterial cultures were performed for four patients with otorrhea, which was controlled by diluted vinegar irrigation with a topical antibiotic solution. The most common bone destruction sites were the inferior and posterior walls. All patients required surgical treatment. Four patients (patient nos. 1, 3, 4, and 5) were treated via a postauricular transcanal approach. Three patients (patient nos. 2, 6, and 7) required mastoidectomy.

Conclusion: Pediatric EACC is not less aggressive than adult EACC. Therefore, early diagnosis and adequate treatment are necessary. Further studies are required to elucidate the clinical features of pediatric spontaneous EACC.

1. Introduction

A cholesteatoma mostly occurs in the middle ear and is rarely found in the external auditory canal. External auditory canal cholesteatoma (EACC) is a rare entity with an estimated occurrence of one in 1000 new patients visiting otolaryngology clinics [1]. EACC occurs mostly in the elderly, and pediatric EACC is very rare. To date, only 21 cases (6 papers) of pediatric EACC have been reported in the literature [2–7].

EACC is caused by an invasion of squamous tissue into a localized area of periosteitis in the bony canal wall [8]. The exact etiology and pathophysiology of EACC has not been elucidated [9]. Patients with EACC typically present with otorrhea and a chronic, dull pain due to the local invasion of squamous tissue into the bony EAC [8]. Generally, middle ear cholesteatoma in children is considered more aggressive than that seen in adults. However, the clinical characteristics of pediatric EACC remain unknown because of its rare occurrence. To date, only Kim et al. [6] have shown that pediatric EACC has a less aggressive growth pattern compared to adult EACC. Further studies are required to understand its clinical behavior, i.e., its aggressiveness. Recently, we encountered seven cases of spontaneous pediatric EACC. The purpose of this study was to evaluate the clinical characteristics of pediatric EACC.

2. Materials and methods

The clinical records of all patients diagnosed with EACC in our department from January 1, 2012 to February 29, 2016 were retrospectively reviewed, focusing on the extension of bone erosion, symptoms, and clinical findings. This study was approved

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Table 1
Staging of spontaneous external auditory canal cholesteatoma using a modified version of the staging system proposed by Naim et al. [6].

Stage	Characteristics
Stage I	Hyperplasia and hyperemia of the auditory meatal epithelium
Stage II	Localized inflammation of the hyperproliferated epithelium and adjacent bone erosion
Stage III	Destruction of the bony canal with sequential bone
Stage IV	Spontaneous destruction of adjacent anatomical structures such as the mastoid, skull base, temporomandibular joint, and facial nerve

by the IRB of Chonnam National University Hospital. EACC was diagnosed by otoendoscopy, otomicroscopy, and temporal bone CT. The location of bone destruction was determined, EACC staging was performed using a system proposed by Naim et al. [6] (Table 1), and treatment outcomes were evaluated. If the bone destruction extended to the skull base or inner ear, temporal bone MRI was performed. Pus culture and antibiotic sensitivity test were routinely performed if otorrhea was detected in the EAC. A pure-tone audiogram was acquired, and if it was not feasible, auditory brainstem response was recorded. The surgical treatment was performed by single senior surgeon (CH Jang).

3. Results

During the study period, eight patients were diagnosed with EACC, but one was excluded because of secondary EACC caused by temporal bone fracture. The remaining seven patients had primary pediatric EACC. Their ages ranged from 5 to 17 years. Table 2 shows their clinical characteristics. All patients showed unilateral EACC. Otalgia and intermittent otorrhea were common symptoms. Bacterial cultures were performed for four patients with otorrhea, which was controlled by diluted vinegar irrigation with a topical antibiotic solution. Bacterial culture test revealed no growth (case no. 2 and 6), MRSA (case no. 4) and *Morganella morganii* subspecies (case no. 7). Three patients were treated using an ototopical ciprofloxacin/dexamethasone solution, and one patient (case no. 4) was treated using an ototopical vancomycin solution because MRSA was identified. The vancomycin solution was fortified with distilled water and vancomycin powder [10]. The most common bone destruction sites were the inferior and posterior walls. All patients required surgical treatment. Four patients (patient nos. 1, 3, 4, and 5) were performed canaloplasty or atticoplasty via a postauricular incision approach. Three patients (patient nos. 2, 6, and 7) required mastoidectomy. In patient no. 3, the mastoid air cells were exposed by posterior wall destruction using a drill. After drilling, the mastoid cavity was investigated by endoscopy. No invaded keratin layer was identified in the mastoid cavity. The bone dust was collected from the surface area of the cortical bone by using a Sheehy bone dust collector (OTOMED, Memphis, USA). The bone defect of the posterior wall was repaired without mastoidectomy via the transcanal approach using bone dust mixed with fibrin glue. In patient no. 5, EACC invaded the attic causing erosion of the scutum. After removing the EACC via the transcanal approach, simultaneous atticoplasty was performed using cartilage. Patient no. 6 had an infected keratin mass with granulation in the EAC. The otorrhea was treated using diluted vinegar irrigation with topical ciprofloxacin/dexamethasone. After the EACC had dried, otoendoscopy was performed, which revealed automastoidectomy. The granulation tissue was identified at the site of a labyrinthine fistula in the

Table 2
Characteristics of pediatric EACC.

Patient no.	Age	Sex	Site	Sx	BD site	Stage	Treatment	FU mo
1	5	M	Lt	ear fullness	inf	III	CP & ICWR	39
2.	14	F	Rt	otorrhea	post	IV	CWUMT1 & PCWR	27
3.	14	F	Lt	no	post	III	CP	18
4.	7	M	Rt	otorrhea	inf	III	CR	18
5.	7	M	Lt	no	attic, inf	IV	CP & atticoplasty	17
6.	17	F	Rt	otorrhea	post & LF	IV	modified radical Mx	20
7.	13	F	Rt	otorrhea	post, inf	IV	CWUMT with staging	1

M: male; F: female; Lt: left; Rt: Right; inf: inferior; post: posterior; LF: labyrinthine fistula; CP: canaloplasty; ICWR: inferior canal wall reconstruction, PCWR: posterior canal wall reconstruction; CR: cholesteatoma removal; CWUMT: canal wall-up tympanomastoidectomy. Case no. 6: Trisomy.

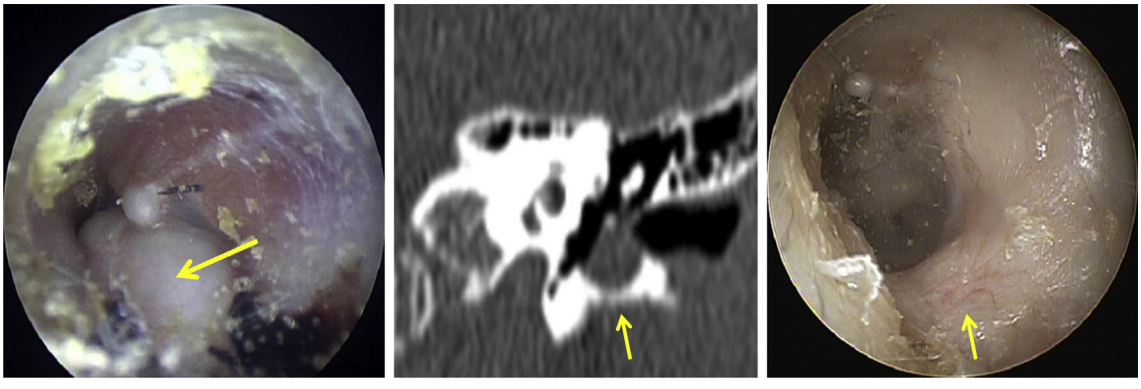

Fig. 1. Patient no. 1. Otoendoscopy shows a white mass (arrow) in the medial side of the external auditory canal (A). CT shows erosion of the inferior canal wall caused by the cholesteatoma (B). Postoperative otoendoscopy shows the inferior wall reconstructed using bone pate with fibrin glue (C).