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CASE REPORT

External Auditory Canal Cholesteatoma

Jacinto Negreiros, Henrique Fernandes Oliveira, Caio Athayde Neves, Carlos Augusto Oliveira

Armed Forces Hospital, Brasilia-DF, Brazil (JN, HFO, CAN)
Department Otolaryngology Brasília University Medical School, Brasilia-DF, Brazil

External auditory canal cholesteatoma is an uncommon disease that is characterized by desquamating tissue and bone erosion in older patients. The lesion could start after a microtrauma in the canal, with consequent squamous epithelium proliferation and periostitis. Another possibility is the loss of the skin migration property, which causes retainment of desquamated cells. It is often associated with infection but it generally presents few symptoms. In this paper we present two cases of external auditory canal cholesteatoma, one of them in a middle age patient, unlike the highest prevalence in older patients.

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External Auditory Canal Cholesteatoma (EACC) is a rare disease, which occurs most frequently among the elderly at an estimated rate of one out of every 1.000 otologic patients.[1,2] It generally presents slow progression and few symptoms, of which chronic otalgy and persistent otorrhea are the most common. Progressive bone destruction typically occurs while the tympanic membrane is spared, but the disease can affect adjacent structures in an advanced stage.[1-3] EACC diagnosis is clinical, aided by imaging and histopathological tests. Differential diagnosis includes keratosis obturans, necrotizing external otitis and tumors.[4,5] Treatment is surgical, varying with the lesion extension and progression.[1-3] We present two cases of external auditory canal cholesteatoma and discuss the etiology, diagnosis and treatment of this condition.

Case Report 1

A 31-year-old woman presented with a 15-day-history of intermittent left ear pain, without otorrhea, tinnitus or vertigo. Physical examination - otoscopy - revealed hard cerumen-covered tumor in the left External Auditory Canal (EAC) which prevented proper tympanic membrane visualization and caused pain during manipulation. It was then requested Temporal Bone Computerized Tomography (CT), which turned out to show soft-tissue mass occupying the whole length of the EAC, with signs of bone erosion in the inferior wall; normal middle ear with intact articulated ossicular chain and mastoid air cells (Figure 1). Transcanal approach excisional biopsy, under local anesthesia, was performed with the help of a surgical microscope resulting in complete removal of the lesion, including skin fragments. It was made using lidocaine 2% and adrenaline 1:80.000U injected under conduct skin. The tympanic membrane was intact and translucent. There was desquamating tissue on friable skin and exposed areas and bone erosion. Histopathological examination showed EAC cholesteatoma (Figure 2).

The patient improved after surgical intervention, presenting good canal skin cicatrization, and has been under observation by our service for 6 months with the use of otomiscroscopy, audiometry and CT exams, which have not indicated lesion recurrence.

Case Report 2

A 79-year-old man with previous hypoacusis presented with a 2-month-history of mild left ear pain and aural fullness, without chronic ear inflammation symptoms. Otoscopy revealed desquamating infected tumor and cerumen occupying the left EAC, which were removed surgically under otomicroscopy and local

Corresponding address:
Caio Athayde Neves
Est Contorno do Bosque s/n Hospital das Forças Armadas - Otorrinolaringologia - Brasília - DF - Brazil 70658-800
Phone: 5506139662350; Fax: 5506132330812; E-mail: caioath@yahoo.com.br;

anesthesia, as described above. It was noted bone erosion in inferior wall of EAC although the tympanic membrane was intact.

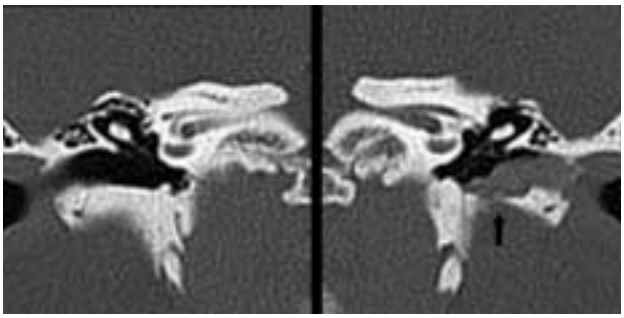

Figure 1. Axial temporal bone CT of patient 1: soft-tissue mass occupying the whole length of the left EAC, with signs of bone erosion in the inferior wall (arrow).

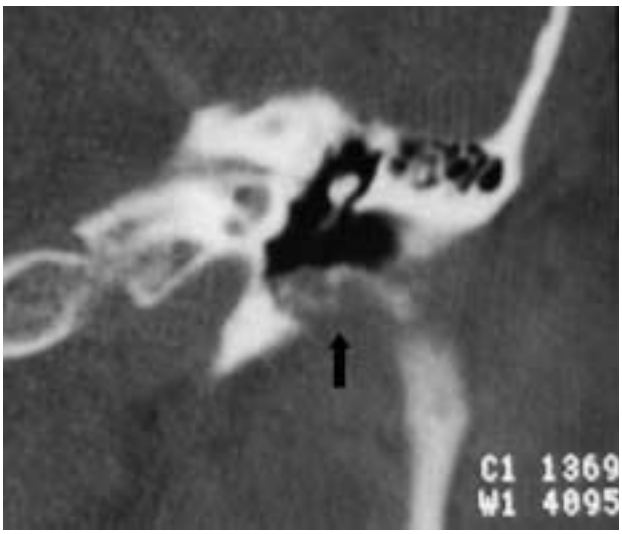

Figure 2. Axial temporal bone CT of patient 2 (left ear): bone erosion in the inferior wall (arrow).

Temporal bone CT showed normal middle ear and ossicular chain and signs of bone erosion in the inferior wall of left tympanic bone (Figure 3). Histopathological diagnosis was external auditory canal cholesteatoma. The patient was improved after the lesion excision, and had no recurrence signal after 9 months.

Discussion

EACC occurs most frequently among the elderly despite cases reported in children and young adults, such as the first patient.[6] Most EACC are acquired, but the first report could be congenital since this patient is not elderly. It presents unilateral occurrence in healthy people, although synchronous bilateral occurrence1 has been reported, as well as cases in immunocompromised patients.[7] The most common symptoms are chronic otalgy and persistent otorrhea, which can be fetid due to secondary infection.[3] Conductive hearing loss may occur in case of canal obstruction. Thus, the patient's hearing is usually preserved, presenting, upon otoscopy, normal tympanic membrane and focal lesion in the external auditory canal.[2] This lesion occurs due to localized periostitis with bone sequestration by proteolytic enzymes and it affects the inferior portion of the canal and causes accumulation of squamous tissue.[3] Osteonecrosis and overlying focal loss of epithelium are the most reliable characteristics favoring EACC.[5]

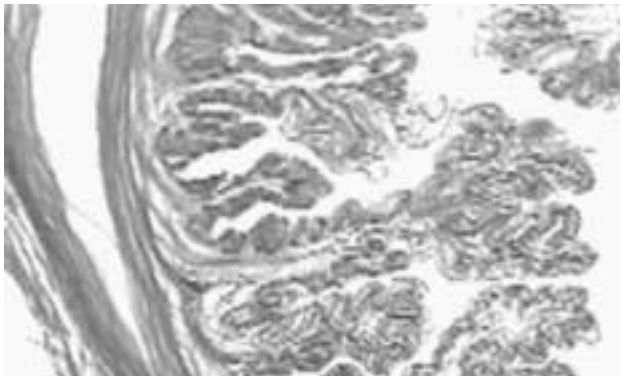

Figure 3. Histological plate of patient 1 - left ear EACC (Hematoxilin and Eosin stain): keratin debris and granulation tissue.

Etiopathology remains uncertain although two mechanisms are pointed as possibilities. The first sustains that the lesion starts with a microtrauma in the canal, followed by periostitis with subsequent invasion and proliferation of squamous epithelium. The second points to possible loss of the skin migration property which causes desquamated cells to be retained by dry cerumen adhered to the canal.[1,3] Such hypotheses explain the appearance of EACC in postoperative situations as well as accompanying canal trauma, canal obstruction by tumor or cerumen and also in young patients.[2,3]

Naim et al classified EACC into four stages: Stage I, canal epithelium hyperplasia; Stage II, presence of periostitis; Stage III, bony canal erosion; Stage IV, erosion of adjacent structures.[8]

Diagnosis is clinical. Computed tomography of mastoid may reveal bone erosion. Histopathologic analysis reveals that Cholesteatoma might be invading neighboring structures. Differential diagnosis must include keratosis obturans, necrotizing external otitis and tumors.[1-3]

Keratosis obturans is a rare condition in which the canal bony portion becomes occluded by desquamated keratin. Its removal reveals canal narrowing, hyperemia with granulation tissue. It occurs bilaterally, among young people, causing acute pain and conductive hearing loss. Imaging of keratosis obturans shows usually bilateral soft-tissue plug in EAC without bone erosion. Its management is clinical.[4,5]

Necrotising external otitis is rare, and basically attack immunocompromised patients and diabetics. Pseudomonas aeruginosa is the most common germ isolated. Severe otalgy, granulation tissue and skin necrosis can be found. Positive culture is important to prescribe a proper antibiotic.

EAC osteoma is a rare benign tumor with 0.05% incidence in patients submitted to otologic surgery. It appears as a small peduncular bony mass starting at the tympanomastoid or tympano-scamous suture, adjacent to the osteocartilaginous junction. It has slow growth, usually asymptomatic, unless there is obstruction of the EAC. It is usually calcified (densely ossified) on CT. There is association with EACC. Treatment is conservative or surgical depending on tumor extension.[9]

Malignant tumors are related with sun exposure. Spinocelular carcinoma is the most common. The lesion is agressive, progressive and frequently multiples. It is infiltrative and have irregular limits. Surgery is essencial, and could be necessary radiotherapy.

Surgical treatment is recommended for EACC, especially in case of chronic pain, constant infection and complications (hypoacusis, facial paralysis, chronic vertigo, lesion progression, hypotympanum involvement, jugular foramen or mastoid involvement). It consists of cholesteatoma matrix and necrotic bone removal.[2] The exposed area must be covered with temporalis muscle fascia graft. Modified radical mastoidectomy must be performed if mastoid cells are affected, with removal of meatus posterior wall and preservation of tympanic membrane and ossicles.[3]

Conclusion

The cases presented here illustrate EACC clinical presentation, diagnosis radiologic charactheristics, treatment and histopathologic features. The occurrence of EACC before old age is unusual. In spite of be a benign lesion, differential diagnosis must be done, and the histopathological examination is essencial to exclud tumors.

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