

Case Report**Recurrent Ear Discharge in Primary Care: Exploring the Possibility of Cholesteatoma - A Case Report**Aneesa Abdul Rashid ^{1,2*}, Khadijah Mohd Nor ^{3,4}, Loh Tze Liang ³Muhammad Farid Hashim ², Faezah Hassan ^{1,2}**Abstract**

Cholesteatoma is a benign lesion of the keratin-producing squamous epithelium, most commonly arising in the middle ear and mastoid. It is rarely found within the external auditory canal. It presents mainly as unilateral lesions and is often destructive and locally invasive. Hence, early detection and referral to an otorhinolaryngology specialist are warranted due to its grave sequelae. This report is about a rare and atypical case of an external ear canal cholesteatoma (EECC) in a young patient. The diagnosis and referral were delayed due to the nature of the disease that resembles an infection, the rarity and atypical presentation of the case, and a low index of suspicion. Therefore, a high index of suspicion based may lead to a timely diagnosis, management, prevention of serious complications, and optimal preservation of ear function

Keywords: cholesteatoma, external ear canal cholesteatoma, ear discharge, primary care, general practice

International Journal of Human and Health Sciences Vol. 08 No. 02 April'24
DOI: <http://dx.doi.org/10.31344/ijhhs.v8i2.647>

Introduction

A cholesteatoma is an ear lesion made up of a mass of squamous epithelium which has undergone stratified keratinization. Although the exact cause of cholesteatoma is unknown, it is believed to originate from the tympanic membrane's lateral epithelium and spread into the middle ear as a self-sustaining mass. Due to possible infection of the dead epithelium in the lesion's centre, this could activate local osteoclasts which could have potentially dangerous effects due to local tissue damage.¹ The estimated incidence of external ear canal cholesteatoma (EECC), an uncommon

disease, is 1.2 per 1,000 new otological patients.² The auditory canal becomes clogged with epithelial debris when EECC occurs. Toynbee and Scholefield first described these symptoms in 1850 and 1893, respectively. Although some cases have presented as EECC, it's possible that they were instances of keratosis obturans, a condition with comparable symptoms. While pain and otorrhea are common symptoms, many cases are astonishingly silent or even asymptomatic.² Therefore, EECC might be a deceptive that may hide major harm while exhibiting little to no symptoms, making it easily overlooked in primary care, as illustrated in this case.

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Case Report

A 22-year-old female presented to a primary care clinic with a complaint of a persistent right ear discharge for a week. The discharge was non foul smelling with some blood streaks and was initially associated with pain, which later resolved. She had received a course of Amoxicillin 500mg three times a day, for one week from a general practitioner (GP). Nevertheless, her ear discharge did not resolve. She has a history of recurrent right ear discharge which was resolved with medication. There was no associated headache, vertigo, facial asymmetry, cough, fever, reduced hearing, or any nasal symptoms. She also denied any history of recent trauma, fall, recent swimming in a pool or river. She neither has known medical illness, allergy, nor past surgical history. She does not smoke or consume alcohol.

On examination, she was comfortable, alert, pink, and not septic-looking. Her body temperature was 37 °C, her blood pressure was 116/65 mmHg, her pulse rate was 103, regularly regular with good volume, and her body mass index was 20.6 kg/m². Examination of the right ear showed the absence of swelling, erythema, and tenderness of the retro auricular region. Otoscopic examination of the right ear revealed mucoid yellowish and whitish discharge with an obscured tympanic membrane (TM). The left ear was normal. Other systemic examinations were unremarkable. Her provisional diagnosis was unresolving right otitis media/right suppurative/exudative otitis externa, and she was referred to the Otorhinolaryngology (ORL) team on the same day.

The otoscopic examination by the ORL team showed granulation tissue at the posterior canal wall with surrounding whitish keratin pearl debris and mucoid discharge, which is typical of cholesteatoma. The TM was not visualised (see Fig. 1). Following the right ear toileting, granulation was seen coming from posterior canal wall and more keratin debris seen. The TM was intact, and the ear canal appeared widened (see Fig. 2). The left ear was noted to be covered with dry keratin material, with a widened ear canal. The left-ear toilet was also done. The provisional diagnosis at this point was right ear canal cholesteatoma with secondary infection.

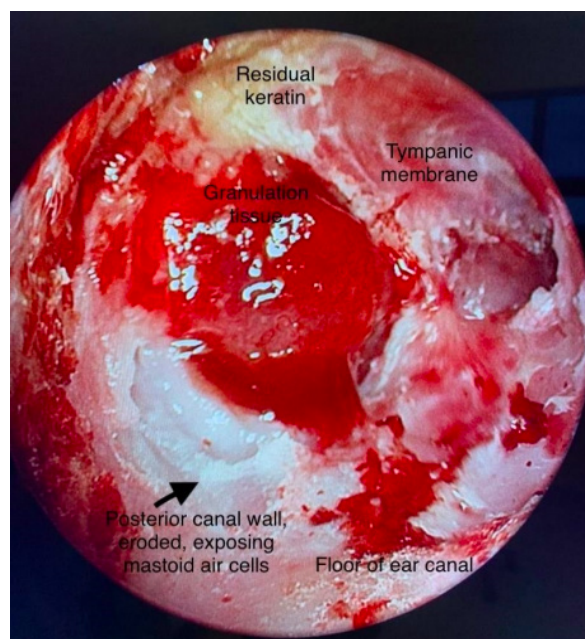


Figure 1: Otoscopic ear examination for first visit



Figure 2: Otoscopic examination after ear toileting of first visit

She was prescribed oral cefuroxime 500mg twice a day and ofloxacin ear drops, five drops twice a day, for a week to treat her infection.

She came back a week later with reduced right ear discharge. There was no ear pain or reduced hearing. Otoscopic examination of the right ear demonstrated a defect at the posterior wall with smaller granulation tissue, and the TM was intact. Pure-tone audiometry performed the same day showed normal bilateral hearing. She was scheduled for an urgent high-resolution computed

tomography (HRCT) of the temporal bone in view of suspected extension of the cholesteatoma into the mastoid cavity via a defect of the posterior canal wall. She was also started on another course of oral antibiotics to treat a superimposed infection.

The findings of the HRCT showed minimal soft tissue density in both external auditory canals, which was likely keratin debris. The scutum was intact bilaterally, with no soft tissue seen in the Prussak space. No fluid or soft density was noted within either middle ear cavity. Tegmen tympani are intact, while the incudo-malleolar complexes are preserved. The internal auditory and facial nerve canals were normal bilaterally. However, fluid density was noted within both mastoid air cells with sclerosis of the right mastoid process. The rest of the visualised paranasal sinuses are clear. There were no bony erosions. The final impression was fluid within both mastoid air cells, and sclerosis of the right mastoid process which may represent mastoiditis. There were no CT features of middle ear cholesteatoma.

An otoscopic examination on her third visit two weeks later showed that the external ear canal (EAC) was expanded and there was opacification of the right mastoid air cells, suggesting previous bony erosion leading to bony remodelling and residual inflammatory or healing processes (see Fig. 3).

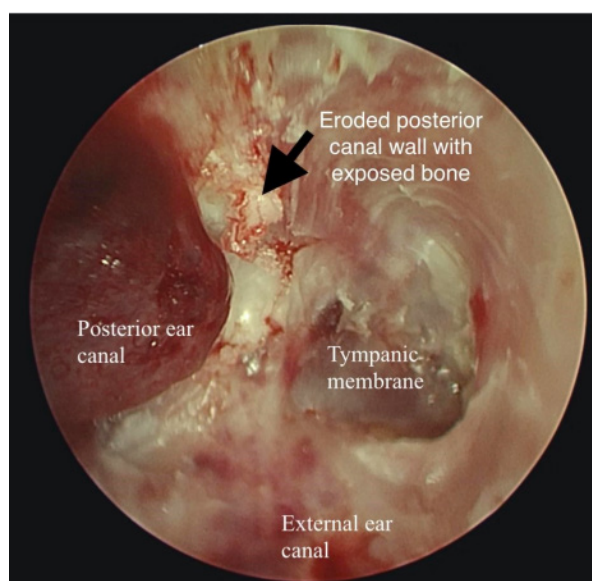


Figure 3: Two weeks post ear toileting visit
The HRCT scan done one month after her first visit neither showed any residual lesion within the

EAC, nor any extension into the mastoid cavity (see Fig. 4). Hence, the final diagnosis of right EECC without mastoid extension was made based on the clinical findings and the good response towards the management.

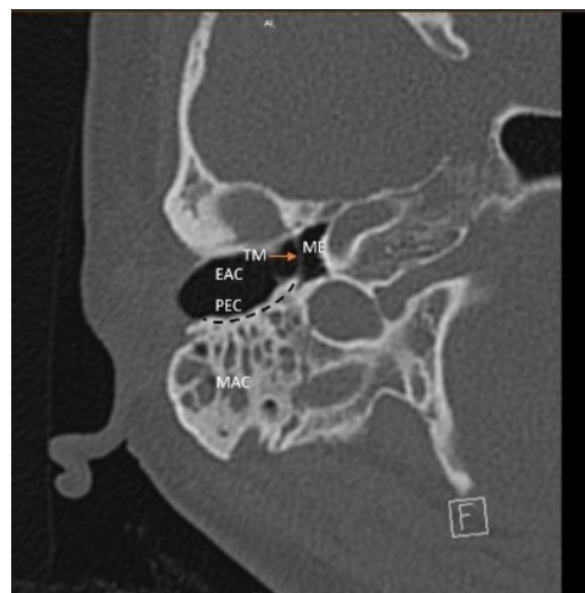


Figure 4: HRCT scan done one month after first visit

After a year of monitoring, she developed a right ear discharge with keratin debris but responded well to ear cleaning and a course of antibiotics. Now, after two years, her ear is dry with normal hearing. An otoscopic examination showed minimal keratin debris, which was cleaned easily. The TM was intact, while the focal mastoid defect was clearly visualised with no keratin within. Accordingly, her follow-up was reduced to twice a year to monitor for recurrence. She was advised to come back immediately whenever she gets another episode of ear discharge.

Discussion

We report a unique case of cholesteatoma, as it is typically located in the middle ear, behind the eardrum.³ Instead, our patient had cholesteatoma in the external ear canal (EECC). The age of presentation is usually in the older age group, in contrast to our young patient in her early twenties in which congenital cholesteatoma may be more likely.⁴ The exact aetiology and pathophysiology of cholesteatoma have not been strongly established.³ This makes cases such as this challenging particularly in the primary care setting.

As illustrated in this case, EECC is primarily diagnosed clinically. As mentioned earlier, this case report is a unique presentation as EECC is classically seen in elderly and has no hearing loss. Nevertheless, there has been reports to debunk the age factor.⁵ Another feature that about one fifth of patients have are a group of symptoms of otorrhea, tinnitus and hearing impairment, was not present in this patient.³ Having said this, this case also showed the usual symptoms of pain and otorrhea in EECC.³

In this patient, an urgent HRCT scan was done, and there were no signs of erosion into the mastoid bone. Most likely because of early detection of EECC. A HRCT was to classify the disease for further management, mainly ruling out the destruction of the nearby temporomandibular joint, mastoid cell, tympanic membrane, or tympanic cavity.⁶

Bilateral EECC has been described in previous literature and a patient may present with unilateral EECC initially with contralateral ear only being involved later.⁷ This is consistent with the findings in our patient whereby the contralateral ear also shows evidence of a widened posterior canal wall which may have been due to spontaneous resolution of EECC.

Management of EECC depends on its growth. Disease limited to the external ear canal requires careful repeated ear toileting to completely remove the cholesteatoma sac followed by ear care with topical or oral antibiotics to resolve localised infection.⁶ Interval follow-up is recommended to assess for resolution of the disease as well as to monitor for recurrence.⁶ As done in this case HRCT temporal is necessary to assess extension of cholesteatoma and if needed, it's appropriate surgical treatment plan.⁸ Depending on the crusting conditions and infection, treatment may be canaloplasty or tympanoplasty.⁶

Here is a concise overview of the primary considerations in the differential diagnosis of canal cholesteatoma (Table 1). While all these diagnoses may present with chronic ear discharge, there are notable distinctions among them.

The key differential diagnosis mentioned above highlight the importance of integrating the clinical, radiological, and histological findings for accurate

Table 1: Differential Diagnosis of Canal Cholesteatoma.

Differential diagnosis	Clinical presentation	Investigation
Keratositis obturans	Presents with bilateral ear canal obstruction and otalgia.	No focal bony erosion.
Chronic otitis externa	Trauma burns or radiation. Granulation and oedema of canal skin.	No bony erosion on imaging.
Post-inflammatory medial canal fibrosis	History of inflammation.	No bony erosion on imaging.
Necrotizing otitis externa	Seen in elderly diabetic patients. Severe otalgia, granulation tissue and cranial nerve palsies.	Positive technetium scan.
Squamous cell carcinoma or neoplasm of EAC	Otalgia, granulation tissue and cranial nerve palsies.	Does not invade bone like cholesteatoma unless in late stage. Requires biopsy to differentiate from cholesteatoma. Lack of maturation and keratin pearl.

differentiation of canal cholesteatoma from other ear canal lesions. Keratositis obturans is the most commonly discussed.⁹

In this case, the diagnosis was delayed. Henceforward, we would like to highlight that a patient who presented with these conditions in primary care justifies a referral to the ORL specialist; (i) the TM cannot be visualized and no clinical improvement despite antibiotic drops (ii) the edge of the TM was not seen when there is a perforation (iii) the view of the TM is obstructed by a granulation tissue and lastly (iv) presence of a conductive hearing loss with a persistent foul-smelling discharge. However, deterioration of the patient's clinical condition, facial nerve weakness, vestibular dysfunction and severe pain requires an urgent ORL referral for evaluation.

Conclusion

Cholesteatoma remains a challenging entity to manage in primary care despite its severe consequences. Henceforth, several features of cholesteatoma is pertinent to preserve hearing and

prevent severe complications through an early detection and referral.

Source of fund (if any) : none

Conflict of interest: none

Ethical clearance: A verbal consent was obtained from the patient for this publication.

Authors' contribution: AAR, MFH, LTL and KMN treated the patient. FH wrote the initial draft of the article. All authors contributed to the write up and discussion of the case manuscript. AAR wrote the final draft. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript

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