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Diagnosis and management of external auditory canal cholesteatoma (EAC)

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Abstract

Introduction: Cholesteatoma is a lesion formed from keratinized stratified squamous epithelium that occurs in the temporal bone and is often found in the middle ear. Cholesteatoma of the ear canal, commonly referred to as External Auditory Canal Cholesteatoma (EAC), involves the invasion of squamous epithelial tissue into the bone of the external auditory canal, which is locally aggressive and erodes the bone. Other terms introduced by experts include keratoma, squamous epitheliosis, cholesteatosis, epidermoid cholesteatoma, epidermoid cyst, and epidermosis. Management of external auditory canal cholesteatoma can be either conservative or surgical. Conservative treatment is indicated when the cholesteatoma and bone erosion are limited to the external auditory canal. Surgical intervention is required when there is extension to the middle ear or mastoid, the presence of complications or potential complications, persistent otorrhea not responsive to conservative therapy, or significant hearing loss.

Case Report: We report a case of a 21-year-old woman with External Auditory Canal Cholesteatoma (EAC).

Conclusion: The definitive diagnosis of cholesteatoma is made through histopathological examination revealing benign keratinized squamous cells. Extirpationf of granulation tissue and cholesteatoma via a transcanal approach was performed, involving the removal of debris and keratin through surgery. The pathological tissue was scraped until normal skin margins were achieved. Eroded bone was cleaned, and granulation tissue was removed until healthy tissue was identified to facilitate healing.

Keywords: Cholesteatoma; Histopathological; Transcanal**;** Extirpation

1. Introduction

Cholesteatoma is a cystic lesion (mass) bounded by keratinized stratified squamous epithelium and containing keratin debris found in the temporal bone (middle ear, petrous apex, and external auditory canal)[1,2]. Cholesteatoma of the external auditory canal is characterized by the invasion of squamous epithelial tissue into the bone of the external auditory canal and is locally aggressive.[8] External auditory canal cholesteatoma is a rare condition, with an incidence estimated at 0.1% to 0.5% of all ear diseases. The average incidence is estimated to be 1.2 to 3.7 per 1,000 otology patients. This condition is more commonly found in older patients compared to younger individuals, with the most affected age group being between 40 and 60 years[3,4,5].

1.1. Pathophysiology

The etiology of external auditory canal cholesteatoma remains unclear. One hypothesis suggests that it is a reactive process resulting from osteitis. Mechanical factors, such as the use of cotton buds and hearing aids, can lead to stenosis of the external canal, which may occur post-traumatically or post-inflammation, trapping the epithelium and resulting

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in cholesteatoma formation. Inflammation of the skin can also cause stenosis or atresia of the ear canal. The skin epithelium in the external auditory canal is a cul-de-sac area, so if solid cerumen remains in the canal for an extended period, the epithelium located medial to the cerumen may become trapped, leading to cholesteatoma formation. External auditory canal cholesteatoma can occur due to occlusion or stenosis of the external canal, which results in the retention of squamous epithelial debris in the medial portion of the canal that should normally be expelled but is obstructed by the occlusion or stenosis[4,6,7].

1.2. Classification

There are several classifications of external auditory canal cholesteatoma:

Table 1 Classification Based on CT Scan of the Mastoid[7,10].

Degrees	Definition	Management
I	Limited to EAC	Conservative management and Canaloplasty
II	Invasion of the Tympanic Membrane and Middle Ear	Canaloplasty and Tympanoplasty
III	Defect in EAC Involving Mastoid Air Cells	Canaloplasty + Mastidectomy + Tympanoplasty + Reconstruction of EAC
IV	Lesion Invading Beyond the Temporal Bone	Surgical Intervention

EAC: External Auditory Canal

Table 2 Naim Classfication[2,3].

No	Staging	Derajat
1	I	Hyperplasia and hyperemia of the epithelium of the external auditory canal (EAC). Increased apoptosis rate on the surface of the cholesteatoma.
2	II	**Findings:** Local inflammation of the hyperproliferative epithelium with signs of periostitis. No destruction of the EAC bone. Accumulation of keratin debris. Clinically, patients may report dull pain and otorrhea if secondary infection occurs. **a.** Intact epithelial surface, with no visible bone of the EAC. **b.** Epithelial defect present, with EAC bone visible.
3	III	Destruction of the EAC bone with sequestration (aseptic osteonecrosis). Epithelial defect extends to the bone. Accumulation of keratin debris with secondary infection and otorrhea.
4	IV	Spontaneous destruction of adjacent structures (according to subclass) accompanied by otorrhea, hearing loss, facial nerve paresis, sigmoid sinus thrombosis, and intracranial abscess. Subclasses: M: Mastoid subclass S: Skull base and sigmoid sinus subclass J: Temporomandibular joint subclass F: Facial nerve subclass

1.3. Diagnosis

The diagnosis of external auditory canal cholesteatoma is established based on the patient's history, physical examination, and supplementary tests.Patients commonly report otalgia (ear pain) that ranges from mild to severe. Other symptoms include otorrhea (discharge from the ear), which is often purulent, particularly in cases of secondary infection. Some cases may be asymptomatic or present with a sensation of fullness in the ear. Physical Examination[2,4,5] Otoscopy: Examination may reveal the accumulation of keratin debris and cholesteatoma in the ear canal. The tympanic

membrane typically appears normal unless the cholesteatoma has extended into the middle ear. Hearing and Balance Tests, These tests are necessary to assess the degree of extension and destruction caused by the cholesteatoma. Pure tone audiometry may show normal results or mild hearing loss[11]. CT Scan: Useful for visualizing the extent of the disease and planning surgical intervention if needed. The CT scan may reveal a soft tissue mass in the external auditory canal and bone destruction in that area. Necrosis and periostitis appear as irregular bone erosion, and fragments of bone within the cholesteatoma (intramural) may also be observed[13]. Histopathological Examination Conducted to confirm the presence of cholesteatoma and rule out the possibility of malignancy in the external auditory canal. Histopathologically, cholesteatoma is characterized as a cyst of benign keratinized squamous cells, consisting of three components: cystic, matrix, and perimatrix. The cystic component is made up of keratinized squamous cell differentiation, while the matrix is composed of keratinized squamous epithelium resembling cyst structure[12,14].

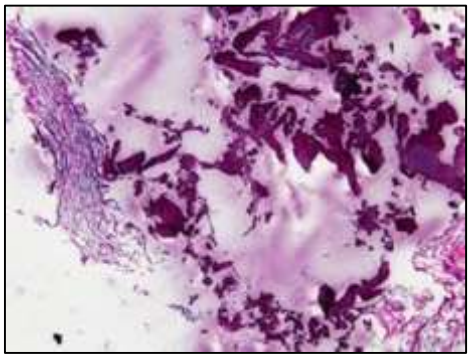

Figure 1 Description: Layered squamous epithelium and subepithelial connective tissue stroma with inflammatory cells[14].

1.4. Management

The management of external auditory canal cholesteatoma can be either conservative or surgical. Conservative Management, This approach is taken when the cholesteatoma and bone erosion are still limited to the external auditory canal. The treatment involves the removal of the cholesteatoma and curettage of necrotic tissue until clean. Tampons are placed and changed periodically until healing occurs, along with the application of topical antibiotics. The goal of this therapy is to control infection and stop drainage from the ear[3,9,15].

Surgical Management: Surgical intervention is indicated when there is extension to the middle ear or mastoid, the presence of complications or potential complications, persistent otorrhea unresponsive to conservative treatment, or significant hearing loss. The surgical principles include cleaning pathological tissue, restoring or preserving hearing function, and maintaining normal anatomical appearance. Stage I: Removal of keratin debris and local treatment with corticosteroids and salicylates. Stages II and III: Surgical removal of debris and keratin. Stage IV: Radical excision of damaged and infected tissue. Surgical Approaches: For Stage I, a transcanal approach is used; for Stages II and III, an endaural approach; and for Stage IV, a retroauricular incision with canal wall down technique[3,8,15].

1.5. Complications

Complications of external auditory canal cholesteatoma correspond to the extent of destruction. These may include, Erosion of the auditory ossicles, Labyrinthine fistula, Facial nerve paresis.

These complications are often the result of extensive extension to the mastoid. Anterior extension can lead to destruction of the temporomandibular joint. Conductive hearing loss may occur if there is obliteration of the external auditory canal[16].

2. Case Report

A 21-year-old female student, referred to as ANG, from Denpasar, presented to the ENT clinic at Wangaya Hospital on September 14, 2024. She was referred from Bhakti Rahayu Hospital with complaints of ear pain radiating to the left jaw for approximately 3 days. At Bhakti Rahayu Hospital, she was evaluated and treated but was informed of a blockage in the left ear. She received ear drops to soften the blockage, but her condition did not improve, leading to her referral for further management.

She reported discharge from the left ear, a sensation of fullness, and hearing loss in that ear. There were no episodes of vertigo, severe headaches, or nausea/vomiting. There was no history of prior ear surgery. The patient had a history of frequently digging into her ears for over 7 years, with instances of bleeding from the left ear.

Figure 2 Examination of the Left Ear

The patient's general status showed good general condition, blood pressure 110/70 mmHg, pulse 84x/minute, respiration 18x/minute, axillary temperature 36.5ºC. Local status Ear Nose Throat Head Neck found no abnormalities in the right ear, no tragus tenderness, no earlobe pulling pain. Left Ear External auditory canal difficult to evaluate due to granulation tissue covering the canal, presence of secretions noted, tympanic membrane evaluation was challenging. In the right ear there no abnormalities in the auricle, no tenderness on tragus palpation, no pain when pulling the auricle, external auditory canal is clear, no discharge, tympanic membrane intact with a visible light reflex, no abnormalities were found on the nose and throat examination. A tuning fork examination was performed with a negative Rinne test result in the left ear (suggestive of conductive hearing loss), a Weber test with lateralization results to the left ear. The patient was diagnosed with granulation tissue in the right external auditory canal and an ear swab examination for culture and CT scan were performed. The patient was diagnosed with external auditory canal cholesteatoma. The patient was treated with Ciprofloxacin 500 mg, twice daily (2 x 500 mg), Paracetamol 500 mg, three times daily (3 x 500 mg) as needed for pain, Ear Drops (Ofloxacin): 3 mg, two drops in the affected ear, twice daily.

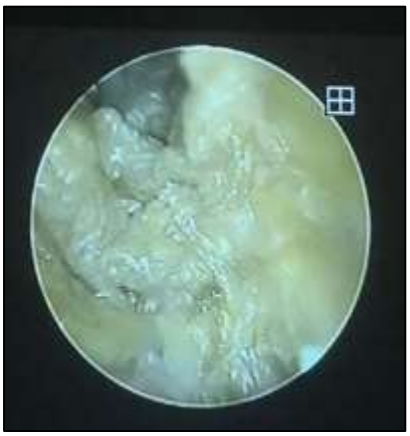

Figure 3 Endoscopic Examination of the Left Ear

On October 8, 2024, a tympanomastoidectomy was planned under general anesthesia. Preoperative preparation for surgery was carried out with routine blood laboratory tests and hemostasis function with results within normal limits. Chest X-ray showed no abnormalities. On October 9, 2024, surgery was performed. The patient was positioned supine on the operating table under general anesthesia, the surgical field was disinfected and draped with sterile towels, a reevaluation of the left ear canal revealed findings consistent with cholesteatoma, covered by granulation tissue. The tympanic membrane in the right ear appeared intact, a decision was made to perform the extirpation of granulation tissue in the right ear canal via a transcanal approach, Lidocaine with adrenaline (1:200,000) was infiltrated into the right ear canal, extirpation of granulation tissue was performed, revealing cholesteatoma tissue, granulation, and cerumen in the right external auditory canal. There was destruction of the skin in the posterior canal down to the inferior aspect, with the posterior canal wall being eroded, while the right tympanic membrane remained intact, meatoplasty was performed on the right side, the ear canal was irrigated with antibiotics, and antibiotic packing was

placed in the right ear canal, pathological anatomy examination was ordered for the surgical findings.After surgery, Postoperative care the patient was prescribed Ceftriaxone 1 g intravenously every 12 hours, Methylprednisolone 31.25 mg intravenously every 2 hours, Tremenza (tizanidine) 60 mg orally every 8 hours.

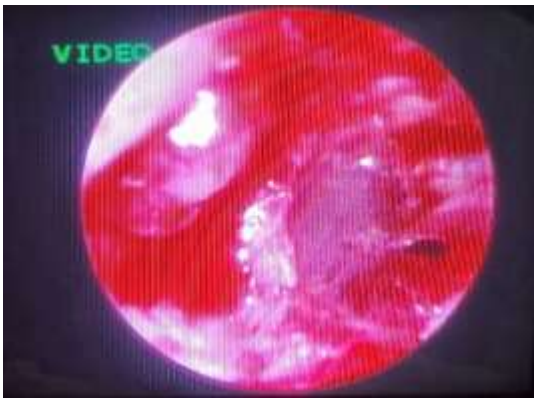

Figure 4 Endoscopic Examination of the Left Ear after extirpation

Follow up one day after surgery, the patient reports mild pain at the surgical site, which is manageable and does not significantly interfere with daily activities, no complaints of dizziness or vertigo noted. The patient was then allowed to go home with ciprofloxacin therapy 500 mg every 12 hours intraorally, paracetamol 500 mg every 8 hours intraorally and methylprednisolone 8 mg every 12 hours intraorally. The patient was advised to keep the ear dry to promote healing, a follow-up appointment is scheduled for one week later to monitor recovery and assess the surgical site.

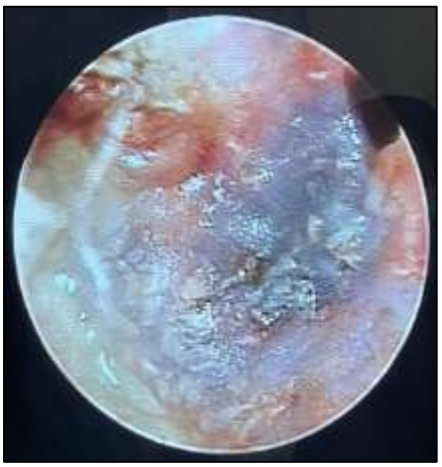

Figure 5 Endoscopic Examination of the Left Ear after 1 day extirpation

On October 16, 2024, The patient returned for a follow-up visit with the pathology results to the ENT-KL polyclinic of Wangaya. The results of the examination showed that the microscopic examination showed tissue samples consisting of keratin material without epithelium. In some areas, fibrous tissue with lymphoplasmacytic inflammatory cells was observed. Conclusion: left external auditory canal, keratin mass, possibly part of a cholesteatoma. the ear tampon in the right ear canal was removed, ear toilet was performed, patient reported no pain, no discharge from the right ear was noted, and hearing in the right ear was reported as improved compared to before the surgery. The patient is advised to continue regular follow-up appointments weekly at the ENT clinic. The next plan includes tympanoplasty.

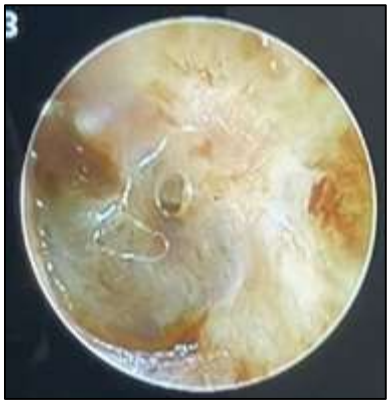

Figure 6 Endoscopic Examination of the Left Ear after 1 week extirpation

During the follow-up at two weeks post-extirpation, the condition of the perforation in the ear canal has improved. The ear pain has also resolved.

Figure 7 The left ear canal of the patient two weeks after extirpation

3. Discussion

Cholesteatoma of the external auditory canal is a rare condition, with an incidence of approximately 0.1-0.5% among new patients presenting with ear problems. The disease progresses slowly and is more common in older individuals. However, some cases have been reported in younger patients, as noted by Indira Acharya and Pabina Rayamajhi, as well as Luciana Almeida Moreira et al. in 2018[5]. In this case, the patient is a young adult, aged 21.

The etiology of cholesteatoma in the external auditory canal remains unclear. However, some experts suggest that it may arise from trauma to the ear canal, chronic inflammation, stenosis of the canal, or even spontaneously[4,6,7]. In this case, the possible etiology includes trauma due to frequent ear cleaning and a history of ear bleeding.

Among 28 patients with cholesteatoma of the external auditory canal reported by Kuen-Yao Ho et al., the most common clinical symptoms were ear pain (19 patients), ear discharge (14 patients), a sensation of fullness (10 patients), tinnitus (5 patients), and hearing loss (4 patients)[3]. In this case, the patient reported left ear pain, discharge, a feeling of fullness, and decreased hearing. Otoscopic examination revealed granulation tissue filling the left ear canal. The diagnosis of cholesteatoma was made intraoperatively when the tympanic membrane was found intact. Definitive diagnosis is confirmed through histopathological examination, which in this case showed keratin mass.

Most reported cases are at least stage IIb, likely due to the difficulty in distinguishing early-stage cholesteatoma from cerumen obstruction or keratosis obstruction[8,14]. This patient was categorized as stage II, as there was no destruction of the ear canal bone.

The management of cholesteatoma in the external auditory canal primarily involves excising the cholesteatoma while preserving the integrity of the ear canal. For stage I, removal of keratin debris and local treatment with corticosteroids

and salicylates are indicated. In stages II and III, surgical extirpation of debris and keratin is performed, with pathological tissue scraped away until healthy tissue is reached. Eroded bone is cleaned, and granulation tissue is removed to facilitate healing. Stage IV requires radical excision of damaged and infected tissue. Ramin N et al. recommend surgical approaches for cholesteatoma in the external auditory canal, including a transcanal approach for stage I, endaural approach for stages II and III, and retroauricular incision with canal wall down technique for stage IV[8,14]. In this case, granulation tissue and cholesteatoma were extirpated via a transcanal approach, along with right meatoplasty under general anesthesia.

4. Conclusion

We reported a case of cholesteatoma in the left external auditory canal of a 21-year-old female patient who underwent successful extirpation of granulation tissue and cholesteatoma via a transcanal approach, with favorable surgical outcomes.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of ethical approval

This study was proposed in ethical clearance application in our institution and hospital, and was already approved to continue with this research. The approval allowed us to interview the patient involved in this study according ton their symptoms

Statement of informed consent

Informed consent was obtained from participants in this study.

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