

Case Report**Management of recurrent chronic suppurative otitis media with cholesteatoma****Finna Christianty, Ahmad Dian Wahyudiono**Department of Otorhinolaryngology Head and Neck Surgery,
Faculty of Medicine Universitas Brawijaya/Dr.Saiful Anwar General Hospital, Malang**ABSTRACT**

Background: Chronic suppurative otitis media (CSOM) is a chronic inflammation involving the middle ear space and it became a burden especially in developing countries. Mastoidectomy remains the main treatment in CSOM with cholesteatoma. Recurrent cholesteatoma and suppurating process indicated the need of revision mastoidectomy. Until now, studies about the mechanism of recurrent cholesteatoma and its management are still scarce. **Purpose:** To analyze factors that played a role in recurrent cholesteatoma and its management. **Case report:** A case of chronic suppurative otitis media with recurrent cholesteatoma which underwent revision mastoidectomy. **Clinical question:** What are the factors associated with the occurrence of recurrent cholesteatoma and how to manage it? **Method:** Evidence base literature study about chronic suppurative otitis media with recurrent cholesteatoma and surgery treatment was performed through PubMed, ProQuest, and Google Scholar databases. **Result:** Recurrent cholesteatoma that occurred after mastoidectomy can be caused by residue of the cholesteatoma itself, or new forming of retraction pocket. Revision mastoidectomy is necessary to evacuate the cholesteatoma and provide good drainage to the mastoid. **Conclusion:** Routine follow-up after mastoidectomy is an important thing to help prevent recurrence of CSOM especially with cholesteatoma.

Keywords: chronic suppurative otitis media, recurrent cholesteatoma, revision mastoidectomy

ABSTRAK

Latar belakang: Otitis media supuratif kronis (OMSK) merupakan suatu kondisi inflamasi kronis pada mukosa telinga tengah dan merupakan masalah kesehatan terutama pada negara berkembang. Pada kasus OMSK dengan kolesteatoma, mastoidektomi merupakan terapi utama. Adanya kolesteatoma rekuren, serta kondisi supurasi, merupakan indikasi diperlukannya mastoidektomi revisi. Hingga saat ini, masih diperlukan pengetahuan yang mendalam mengenai mekanisme terjadinya kolesteatoma rekuren serta talaksanaannya. **Tujuan:** Untuk menganalisis faktor yang berperan pada kolesteatoma rekuren serta penatalaksanaannya. **Laporan kasus:** Satu kasus OMSK dengan kolesteatoma rekuren yang menjalani operasi mastoidektomi revisi. **Pertanyaan klinis:** Apa saja faktor penyebab dan bagaimana tatalaksana OMSK dengan kolesteatoma rekuren? **Metode:** Telaah literatur berbasis bukti melalui pencarian PubMed, ProQuest, dan Google Scholar dengan kata kunci "recurrent cholesteatoma" DAN "surgery treatment." **Hasil:** Kolesteatoma rekuren dapat terjadi pasca dilakukan mastoidektomi akibat adanya residu, atau terbentuknya kembali kantong retraksi. Mastoidektomi revisi diperlukan untuk membersihkan jaringan kolesteatoma dan memperbaiki drainase dari mastoid. **Kesimpulan:** Pemantauan yang ketat pasca dilakukan mastoidektomi adalah hal yang penting untuk mencegah rekurensi dari OMSK, terutama OMSK dengan kolesteatoma.

Kata kunci: otitis media supuratif kronis, kolesteatoma rekuren, mastoidektomi revisi

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INTRODUCTION

The term otitis media refers to a group of disorders in the middle ear resulting from infection and inflammation. It is estimated that as many as 80% of children worldwide have experienced an episode of otitis media. Otitis media is generally subdivided into 2 types, acute otitis media (AOM) and chronic otitis media, where chronic suppurative otitis media (CSOM) is included.¹ In clinical setting, CSOM is defined as prolonged middle ear infection characterized by the presence of purulent ear discharge through a perforated tympanic membrane for at least 2 months. Furthermore, CSOM is subdivided based on the presence or absence of cholesteatoma as tubotympanic CSOM/safe/benign type, and atticointral CSOM/dangerous/malignant type. The tubotympanic type is characterized by a central tympanic membrane perforation, while the atticointral type is characterized by involvement of the postero-superior part of the middle ear (including the attic, antrum, and mastoid) with marginal or atic perforation of the tympanic membrane. The presence of cholesteatoma and erosion of the ossicles are features of atticointral CSOM.^{2,3}

CSOM is a worldwide health problem with prevalence in Southeast Asia of 7.8% based on World Health Organization (WHO).⁴ According to a study conducted by Desbassarie et al.⁵ at Hasan Sadikin Hospital Bandung in 2011, there were 117 cases of CSOM with 43 of them were complicated CSOM cases. The etiology and pathophysiology of otitis media are multifactorial and overlapping, including impaired ventilation function of the Eustachian tube and infectious process involving biofilm and anaerobic bacteria.^{6,7}

Mastoidectomy remains the mainstay therapy in malignant/atticoantral CSOM. The aim of this procedure is creating a dry and safe ear by removing granulation and pathological tissue in the middle ear, thereby eradicating the infection. The next goal after the infection has been eradicated is the preservation and reconstruction of the remaining hearing function. The canal wall down mastoidectomy (CWD) is performed by opening the mastoid cavity and leave it open by removing the entire posterior meatal wall. This technique is known as radical mastoidectomy in which the entire middle ear cavity along with the attic and mastoid antrum becomes a large, merged cavity. Whereas the canal wall up technique (CWU) is performed by cleaning all pathological tissue while maintaining the posterior meatal, so on the contrary, the mastoid cavity is not left open.⁸

Canal wall reconstruction (CWR) tympanomastoidectomy is a technique that combines CWD and CWU with the purpose of optimizing eradication of cholesteatoma tissue, and providing blockade of the attic, thereby reducing the risk of recurrence and formation of retraction pockets. The CWR technique was introduced by Mercke in 1987 and is performed with mastoid obliteration using several methods such as osteoperiosteal flaps, ceramic alloplasts, bone cements, and costal cartilage.⁹

Recurrent cholesteatoma that crops up after mastoidectomy surgery can occur due to residual cholesteatoma that had not been cleaned during surgery, or the formation of a retraction pocket on tympanic membrane through the ear canal that had been reconstructed previously. The presence of recurrent cholesteatoma can increase

morbidity rate and requires eradication.¹⁰

In a study conducted by Vartiainen¹⁰ in 349 subjects after mastoidectomy, as many as 19 subjects (8.3%) experienced recurrent cholesteatoma in varied timespan between 2-12 years after the first/primary mastoidectomy. Revision mastoidectomy is a term that refers to re-surgery/second look surgery to remove pathological tissue that recurred after primary mastoidectomy. Before performing revision mastoidectomy, several things need to be considered, including re-evaluating the CSOM disorders that were experienced before the primary mastoidectomy, to analyze factors that influenced primary mastoidectomy.¹¹

This case report aimed to further analyze factors that played a role in recurrent cholesteatoma and its management.

CASE REPORT

A 20 years old female with chief complaint of foul-smelling mucopurulent discharge on the left ear with hearing impairment for the last 12 years. She came to ENT clinic Dr. Saiful Anwar General Hospital for consultation. Several years ago, on July 2018, she had severe headache accompanied with decrease consciousness and had undergone surgical drainage for cerebral abscess by a neurosurgeon. Patient was referred to ENT clinic after recovering from the surgery. On October 2018, a modified radical mastoidectomy was performed. Cholesteatoma and granulation tissues were found intra-operatively in the entire mastoid cavity; the malleus was already decomposed, stapes and incus were covered with granulation tissues, attic perforation and adherent tympanic membrane were seen. Modified radical mastoidectomy in this primary mastoidectomy procedure was performed by obliterating the mastoid cavity with fibrous tissue.

Dry ear and hearing improvement occurred 9 months after surgery. Two

years after primary mastoidectomy, patient experienced mucopurulent ear discharge and hearing impairment on the same ear.

Otoscopic examination revealed mucopurulent discharge on the left ear, with granulation and cholesteatoma tissues seen in postero-superior region. (Figure 1).

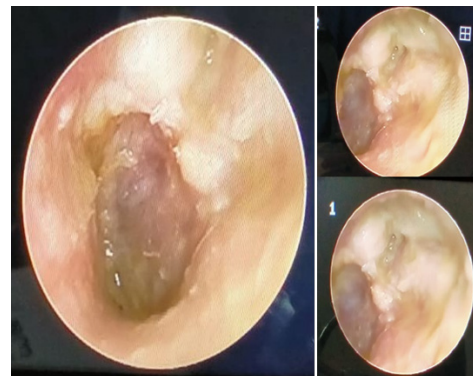


Figure 1. Otoscopic examination 2 years after primary mastoidectomy

Mastoid High Resolution Computed Tomography (HRCT) was performed and revealed: no visible ossicles, with isodense lesions in the mesotympanum, suspected of cholesteatoma. (Figure 2)

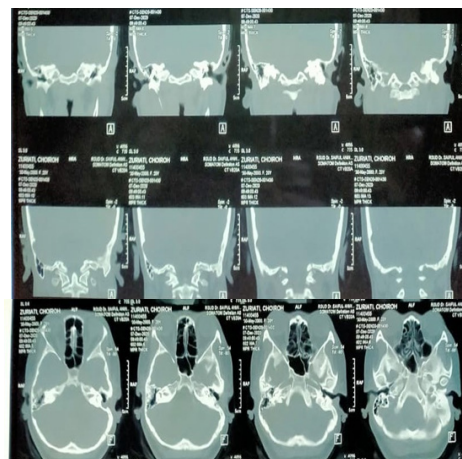


Figure 2. Mastoid HRCT: no visible ossicles, with isodense lesions in the mesotympanum, suspected cholesteatoma

Pure tone audiometry revealed a normal hearing (16.25 dB) on the right ear and mild conductive hearing loss on the left ear (36.25 dB), with 100% speech discrimination on both ears. Tympanometry revealed B type on the left ear.

On January 2021, left revision mastoidectomy with mastoid obliteration was performed. Cholesteatoma which was found on the posterior wall was removed, and mastoid antrum was dilated. Tragal cartilage was harvested as graft material, and used to cover the posterior meatal wall defect (Figure 3).

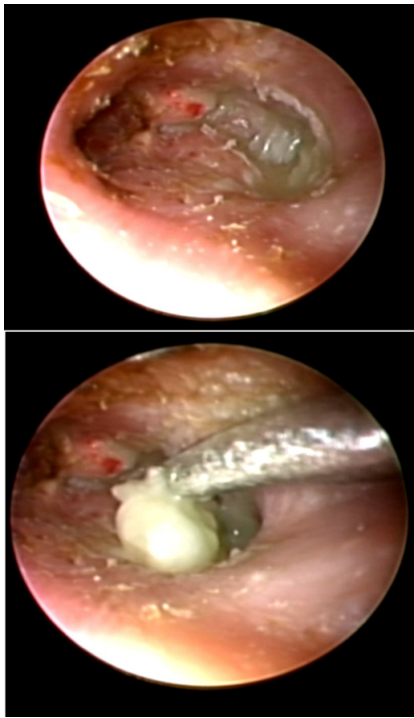


Figure 3. Cholesteatoma tissue on posterior meatal wall

Two months follow-up after revision mastoidectomy surgery, showed a dry ear.

CLINICAL QUESTION

What are the factors associated with the occurrence of recurrent cholesteatoma, and how to manage it?

REVIEW METHOD

A literature search was conducted on February 2021 with the keywords “chronic suppurative otitis media”, AND “recurrent cholesteatoma”, AND “revision mastoidectomy”.

In the PubMed-Medline database, as well as Goggle Scholar, 20 literatures were obtained which were then filtered according to the publications in the last 10 years, relevant with the topic, and available in full text.

RESULT

Based on a study conducted by Vartiainen,¹⁰ recurrent cholesteatoma can occur 2-12 years after primary mastoidectomy had been performed. The recurrence rate of cholesteatoma was found to be different in children and adults; where in children, recurrence after 5 years of primary mastoidectomy was 2.6 times higher than in adults. This was due to the difference in Eustachian tube function, which is better in adults than in children, so the risk of negative pressure in the middle ear is lower.^{10,12}

Risk factors for recurrent cholesteatoma after previous primary mastoidectomy can be divided into pre-operative, intra-operative, and post-operative factors. Pre-operative factors that play a role in the occurrence of recurrent cholesteatoma include age, as well as the presence of previous adhesive otitis media which generally occurs at a young age. This results in prolonged negative pressure, increasing the risk of forming a retraction pocket. Intra-operative factors that play a role in recurrent cholesteatoma include the expansion of the cholesteatoma itself. A cholesteatoma that is large enough to extend to the antrum, tympanic sinus, and mastoid, has a higher risk of leaving residue and recurrence, because it can hinder intra-operative visualization. In addition, the technique related to removing cholesteatoma tissue is also considered as intra-operative factors that is associated with recurrence. Several studies stated that the CWU technique had a greater risk of recurrence than CWD or modified radical mastoidectomy. However, until now, this was considered to have no significant effect because not all similar studies

had the same conclusions. Reconstruction of the posterior meatal wall with a cartilage graft is assumed to play a role in the recurrence of cholesteatoma, because it is thought to 'bury' the pathological tissue in the middle ear. So far, it had not been proven; and on the other hand, reconstruction of the posterior meatal wall will restore the normal anatomy of the external auditory canal.^{12,13}

After mastoidectomy surgery, routine follow-up is required which includes routine otoscopic examinations and ear toilet, as well as tympanogram and audiometry examinations. Examination with otoscopy aims to assess the presence or absence of retraction pockets and cholesteatoma formed post-operatively. In addition, the condition of the tympanic membrane also needs to be evaluated, and in most cases recurrent cholesteatoma is often not accompanied by re-perforation of the tympanic membrane that was previously reconstructed. Type A tympanogram image shows good middle ear ventilation so that the possibility of residual and recurrent cholesteatoma can be ruled out. Yearly post-operative evaluation is recommended for at least 5 years to prevent recurrence.^{12,13}

DISCUSSION

Chronic suppurative otitis media is a prolonged chronic inflammation in the middle ear cavity, which is divided into two subtypes, namely atticotympanic CSOM and tubotympanic CSOM. Surgical therapy with mastoidectomy is the first-line therapy in cases of CSOM with cholesteatoma which aims to eradicate infection, create dry ears, and preserve hearing must be achieved from surgical therapy in CSOM cases: eliminating the progression of the disease to create dry ears, modification of the tympanomastoid anatomy to prevent recurrence of the disease, and reconstruction/preservation of hearing. Failure of achieving one of these goals is an indication for revision mastoidectomy.¹¹

The reported case was CSOM with recurrent cholesteatoma who had previously undergone primary mastoidectomy using a modified radical mastoidectomy technique. Based on several studies conducted by Nadol,¹¹ reported failure of primary mastoidectomy varies between 3%-36%. Factors that play a role in the failure of primary mastoidectomy are varied and multifactorial, including anatomic factors after primary mastoidectomy, to a combination of extension of CSOM itself with the selected surgical technique.^{11,12}

The presence of recurrent cholesteatoma is a sign of failure from previous surgery, and in our patient this condition occurs 2 years after primary mastoidectomy. This is in accordance with a study conducted by Vartiainen¹⁰ where recurrent cholesteatoma can occur 2-12 years after the primary mastoidectomy had been performed.

Factors that associated with recurrent cholesteatoma after primary mastoidectomy can be divided into pre-operative, intra-operative, and post-operative factors, respectively.

In this reported case, evaluation of the adenoid structure was not carried out, so that the recurrence of the disease in the subject might still be caused by Eustachian tube disturbance which could be obstructed by the adenoid. Eustachian tube function is one of the pre-operative factors that play a role in recurrent cholesteatoma, in addition to age, as well as the histopathological condition of the middle ear in CSOM. So, Eustachian tube function test need to be performed. Some theories conclude that, recurrent cholesteatoma is more common in children less than 18 years compared to adults, because of the immaturity of Eustachian tube function. The study conducted by Volgger et al.¹³ concluded that age did not significantly influence the recurrence of cholesteatoma. In the same study, it was surmised that the presence of persistent or known residual

adenoids was one of the independent risk factors influencing cholesteatoma recurrence. The adenoidectomy procedure performed before the primary mastoidectomy had a compelling effect on recurrent cholesteatoma.

Mastoid HRCT performed before revision mastoidectomy revealed isodense lesion indicating a sclerotic process in the mastoid bone. Histopathological factors from the middle ear with CSOM are also important as predictors of mastoidectomy outcome. In chronic infection, deposits of granulation and connective tissues occur in the submucosa, resulting in sequestration of previously well-pneumatized mastoid air cells. In addition, osteoclast activity in the mastoid and ossicles could occur in CSOM. Both conditions can be identified through radiographic examination as poorly pneumatized/sclerotic mastoid bone which appear as opacification on X-ray, or isodense appearance on mastoid HRCT. The presence of this condition needs to be well-understood because it could cause difficulties in identifying surgical landmarks intra-operatively, and increasing the risk of leaving cholesteatoma residues. In the cases described in the study, mastoid HRCT was performed as a routine examination prior to revision mastoidectomy. HRCT examination itself is not a routine follow-up/monitor modality after a mastoidectomy, but it was needed in cases of chronic middle ear infections.^{11,14}

In the cases described in this study, mastoid HRCT was performed as a routine examination prior to revision mastoidectomy. HRCT examination itself is not a routine follow-up/monitor modality after a mastoidectomy, but it was needed in cases of chronic middle ear infections. NO REF MANA??

Mastoid HRCT is currently the main radiological examination in the identification of cholesteatoma tissue. Based on a study conducted by Fathy et al.¹⁵, magnetic resonance imaging (MRI) examination is superior to mastoid HRCT in identifying recurrent cholesteatoma, because the appearance

of cholesteatoma with granulations and secretions will give the same picture on mastoid HRCT examination. While on MRI, cholesteatoma tissue will give a clear hyperintense picture caused by the water molecules in it, which will affect signal diffusion on MRI examination, so it is easier to distinguish from granulation tissue or other inflammatory processes. However, in small cholesteatoma sizes, which are less than 5 mm, HRCT will provide a clearer picture than MRI because small cholesteatoma could possibly be identified as an artefact on MRI examination.

Intra-operative factors that play a role in cholesteatoma recurrence, include the expansion of the cholesteatoma itself, and the selected surgical technique. Extension to the tympanic sinus and antrum is statistically a risk factor for recurrent cholesteatoma. This is because the tympanic sinus is a region that is difficult to reach, and the presence of a cholesteatoma in this region has the risk of expanding vertically and medially towards the facial nerve, making it increasingly difficult to identify.^{10,11}

In our presented case, the findings at the primary mastoidectomy surgery, and also the revision showed the presence of granulation tissue, which could be a risk factor for disease recurrence in patients. According to the study conducted by Merchant et al.,¹⁴ the presence of granulation tissue indicated a defect in the middle ear mucosa due to prolonged negative pressure, thus facilitating the occurrence of middle ear reinfection which results in recurrence.

Primary mastoidectomy surgery in our presented patient was performed with modified radical mastoidectomy where posterior meatal canal was removed. With this technique, it is expected that all previously formed cholesteatoma tissue could be removed optimally, thereby preventing recurrence. Several previous studies revealed that the mastoidectomy

technique with CWU has the risk of leaving cholesteatoma remnants which will lead to recurrence. In a study conducted by Volgger et al.¹⁰, it was found that there was no significant difference between the CWU technique and CWD as a risk factor for recurrent cholesteatoma. In addition, reconstruction of the posterior meatal wall with cartilage grafts is suspected of having a role in the occurrence of recurrent cholesteatoma because it is considered to 'bury' pathological tissue in the middle ear. So far, this had not been proven, and on the other hand, reconstruction of the posterior meatal wall will restore the normal anatomy of the external acoustic canal.^{10,11}

In the revision mastoidectomy that was performed on our reported patient, graft stiffening and obliteration were performed using the tragus cartilage and fascia temporalis. This technique was carried out to prevent the formation of retraction pockets, especially in the attic and was expected to provide better access for post-operative aural toilet. Some modifications that need to be considered in revision mastoidectomy to prevent recurrence are meatoplasty, canaloplasty, and graft stiffening. In the CWU and CWD techniques, an adequate meatoplasty technique by removing a small concave part of the concha cartilage in the posterior part of the external auditory canal can facilitate inspection and cleaning of the ear after the procedure. Canaloplasty is generally performed in cases accompanied by stenosis of the external acoustic canal and has the same goal as meatoplasty. Graft stiffening aims to prevent retraction of the tympanic membrane, and can be achieved by using cartilage from the tragus. The obliteration technique is performed to minimize the size of the cavity formed and create a barrier between the skin and small structures such as the labyrinth and facial nerve. The obliteration technique is expected to be able to prevent the occurrence of granulation tissue and cholesteatoma, especially in the mastoid tip area, the sinodural angle, and the tympanic

tegmen.¹¹

In our reported case, it was found that the patient had a B tympanogram on the left ear that had previously undergone primary mastoidectomy accompanied by otorrhea, so that it could be suspected of recurrence. In addition, the patient also did not perform routine control after primary mastoidectomy. This was a factor that plays a role in the occurrence of recurrent cholesteatoma because post-operative cleaning was not optimal and hearing function was not monitored which could be a sign of recurrence of the disease. Post-operative evaluation is recommended to be carried out for at least 5 years to prevent recurrence.¹¹

Post-mastoidectomy care is very important for good outcome and preventing recurrence. After the procedure, the patient needs to undergo routine ear cleaning (aural toilet) at least twice a week, and can be continued once a week after epithelialization and dry ears are formed, then it can be continued once a month. Granulation tissue formed during post-operative care can be cleaned with cauterization, and the cavity formed can be cleaned with an ear tampon containing an antiseptic liquid such as povidone iodine.¹⁶ In addition, it is necessary to monitor the patient's hearing function with pure tone audiometry and tympanometry. The improvement of the hearing threshold and type A tympanogram after mastoidectomy indicates a small possibility of recurrent cholesteatoma, so that imaging examinations with HRCT mastoid as a consideration for post-operative evaluation can be deferred.^{10,16}

Based on the discussion of the cases above, it can be concluded that the presence of recurrent cholesteatoma is a sign of failure from the previous mastoidectomy procedure which can be caused by pre-operative, intra-operative, and post-operative factors, and can occur within a period of 2-12 years after primary mastoidectomy. Revision mastoidectomy was indicated in our presented

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