

**Case report****Comprehensive management of malignant otitis externa with tuberculosis and cranial nerve paresis in geriatrics****Ratna Dwi Restuti**Department of Otorhinolaryngology Head and Neck Surgery,  
Faculty of Medicine Universitas Indonesia/Dr. Cipto Mangunkusumo Hospital,  
Jakarta**ABSTRACT**

**Background:** Malignant otitis externa is an inflammatory condition of the external ear which has the propensity to spread to the skull base. It can be a difficult entity to treat as clinical presentation varies and response to treatment differs between patients. **Purpose:** Evaluating the management of malignant otitis externa with complications in geriatric patients who had multiple comorbidities. **Case:** A 71 years old female with a diagnosis of left malignant otitis externa with complications of multiple cranial nerve palsies (N.VII, IX, X) and comorbidity in the form of diabetes mellitus and chronic kidney disease. The patient underwent subtotal temporal bone resection and petrosectomy. **Clinical Question:** "Could surgical management of malignant otitis externa with cranial nerve palsies complication in geriatric patients with multiple comorbidities achieve better result than conservative treatment?" **Review Method:** Literature search using keywords 'malignant otitis externa' OR 'temporal bone osteomyelitis' AND 'geriatric' OR 'elderly' AND 'multiple cranial nerve palsy' AND 'diabetes mellitus' AND 'tuberculosis' AND 'surgery' OR 'surgical' was conducted through Cochrane, Pubmed, and Google Scholar. **Result:** The search obtained 11 articles published in the last 10 years. Selection based on inclusion and exclusion criteria, 2 studies were found relevant with the topic. **Conclusion:** Management of malignant otitis externa with complications in geriatric patients with multiple comorbidities requires a multidisciplinary approach to determine the need for surgery intervention.

**Keywords:** malignant otitis externa, cranial nerve palsy, subtotal temporal bone resection, geriatric, diabetes mellitus

**ABSTRAK**

**Latar belakang:** Otitis eksterna maligna adalah suatu kondisi peradangan pada telinga luar yang memiliki kecenderungan untuk meluas hingga ke dasar tengkorak. Penyakit ini menjadi sulit ditangani karena manifestasi klinis yang bervariasi dan respons terhadap pengobatan yang berbeda antara pasien. **Tujuan:** Mengevaluasi tatalaksana otitis eksterna maligna dengan komplikasi pada pasien geriatri yang memiliki komorbiditas multipel. **Kasus:** Pasien perempuan 71 tahun dengan diagnosis otitis eksterna maligna telinga kiri dengan komplikasi paresis saraf kranial multipel (n.VII, IX, X) dan penyakit penyerta berupa diabetes melitus dan gagal ginjal kronik. Pasien menjalani operasi reseksi tulang temporal subtotal dan petrosektomi. **Pertanyaan klinis:** "Apakah tatalaksana pembedahan memberikan hasil yang lebih baik dibandingkan terapi konservatif pada pasien geriatri dengan otitis eksterna maligna disertai paresis saraf kranialis dengan komorbiditas multipel." **Telaah Literatur:** Dilakukan menggunakan kata kunci 'malignant otitis externa' ATAU 'temporal bone osteomyelitis' DAN 'geriatric' ATAU 'elderly' DAN 'multiple cranial nerve palsy' DAN 'diabetes melitus' DAN 'tuberculosis' DAN 'surgery' ATAU 'surgical' pada beberapa sumber data seperti Cochrane, Pubmed, dan Google Scholar. **Hasil:** Didapatkan 11 artikel publikasi 10 tahun terakhir. Berdasarkan kriteria inklusi dan eksklusi diperoleh 2 artikel yang relevan dengan topik. **Kesimpulan:** Tatalaksana otitis eksterna maligna dengan komplikasi pada pasien geriatri dengan komorbiditas multipel, membutuhkan pendekatan multidisiplin terutama untuk menentukan perlunya dilakukan tindakan pembedahan.

**Kata kunci:** *otitis eksterna maligna, paresis saraf kranial, reseksi tulang temporal subtotal geriatri, diabetes melitus*

**Correspondence address:** Ratna Dwi Restuti. Department of Otolaryngology, Head and Neck Surgery, Faculty of Medicine Universitas Indonesia/Dr. Cipto Mangunkusumo Hospital, Jakarta.  
E-mail: ratna.drest@gmail.com.

## INTRODUCTION

Malignant otitis externa (MOE) is an infection of the external auditory canal, which could extend into the temporal bone and the adjacent structures, such as to the mastoid process and the skull base. Spread of infection into the temporal bone occurs through the fissures of Santorini and the tympanomastoid suture, leading to involvement of the stylomastoid and jugular foramens. Venous channels and facial planes facilitate the spread of infection along the dural sinuses, eventually extending to the petrous apex.<sup>1,2</sup>

Osteomyelitis of the temporal bone is not a common entity. Osteomyelitis can be defined as an inflammatory condition of the bone that begins as an infection of the medullary cavity, rapidly involves the haversian systems, and extends to involve the periosteum of the affected area. Osteomyelitis commonly occurs in long bones of the body. The temporal bone is not very often affected. Osteomyelitis of the temporal and adjacent bones was initially described in 1959. It is potentially a fatal infection of the temporal bone that most commonly affects elderly diabetic patients. Osteomyelitis is a complication of otitis externa. Because of the high mortality rate (46%), this condition is often referred to as malignant otitis externa (MOE). The incidence of MOE increases along with patient's awareness to seek medical help, diagnostic technical development, and the escalation number of diabetic and geriatric patients. Geriatric patients are more vulnerable to suffer external ear infection because of their habit to scratch their dry and itchy ears with cotton buds. Besides, diabetes

has caused pH shift in external ear and rises ear infection risk due to autoimmune response change.<sup>3,4</sup>

Severe otalgia and purulent ear discharge are the common symptoms of MOE. On otoscopic examination, the clinical signs are similar to benign otitis externa. However, the presence of edema and granulation tissues at the osseocartilaginous junction in the external auditory canal indicate the existence of MOE. The ability of this disease to spread through the skull base, increases its propensity to present with nerve palsies, the facial nerve (CN.VII) being the most common nerve involved, followed by other cranial nerves (CN. IX,X,XII). This disease is mostly seen in patients with diabetes and immunocompromised status.<sup>1,5</sup>

The most common organism isolated is *Pseudomonas aeruginosa*, and the widely accepted empiric treatment based on clinical studies is 3<sup>rd</sup> generation cephalosporins and fluoroquinolones. It has been noted that some patients do not respond to this treatment and in those cases, fungal MOE due to *Aspergillus* and tuberculosis should be kept in mind. To differentiate the cause, biopsy should be done to rule out the extremely rare case of extrapulmonary TB. Pulmonary tuberculosis (TB) is the most common manifestation of *Mycobacterium tuberculosis*, while tuberculosis of the head and neck area makes up 2-6% of extrapulmonary TB, and 0.1-1% of all forms of TB.<sup>1,6</sup>

The main treatment of MOE is long term antimicrobial therapy. Other treatment strategies are close follow-up of blood glucose levels, repeated local debridement

of necrotic tissues, and hyperbaric oxygen therapy. Surgery has a limited role in the treatment of MOE.<sup>1,7</sup>

Once the patient is symptomatically better, the otolaryngologist faces the dilemma as to when to stop the medications and prevent the chances of recurrence. In this aspect, radionuclide scans i.e. Gallium-67-citrate (<sup>67</sup>Ga) or 111Indium (<sup>111</sup>In) play a significant role as they identify areas of residual infection and these scans return to normal when the infection subsides.<sup>1</sup>

The aim of this case report is to evaluate the management of a difficult case of geriatric MOE patient with clinical manifestation of cranial nerve palsy caused by *M. tuberculosis* infection.

## CASE REPORT

A 71 years old female came with periodical severe pain on the left ear. She had the same complaint 5 months previously and was treated in another town hospital. She had difficulty in swallowing and facial nerve palsy. She had a history of diabetes mellitus of 15 years. Upon examination the outer third of the left ear canal was full of granulation tissue, and mucus discharge was also present. The right tympanic membrane was intact. Facial nerve examination showed left facial nerve palsy with House Brackmann V. Microbial culture and resistance test of the ear discharge found *Pseudomonas aeruginosa*. Hearing function test using *Auditory Steady-State Response* (ASSR) showed severe mixed hearing loss (bone conduction 50 dB and air conduction 70-80 dB). Radiologic examination of the head by *Magnetic Resonance Imaging* (MRI) showed destruction of the left petrous bone, left sigmoid sinus, left mastoid bone, left occipital bone including nasopharyngeal region, external ear canal, left masticator and left mandible angle. Medical Rehabilitation assessment exhibited oral phase dysphagia,

cranial nerve IX and cranial nerve X palsies. The patient also had other comorbid factors, including chronic renal failure, anaemia (Hb 9.2 g/dL), and hypoalbuminemia.

She was given medical treatment for 11 days consisted of Ceftazidime 2x1g IV, Na-Diclofenac and K-Diclofenac 2x50 mg per oral, and Ofloxacin ear drops 2x5 drops on the left ear. She underwent left subtotal temporal bone resection and petrosectomy surgery. Upon surgery, there was pale granulation tissue filling the mastoid tip up to the temporomandibular joint. Intraoperative biopsy revealed caseous necrosis granuloma. Facial nerve sheet was impaired and there was pus producing abscess inside. The dura mater was torn during operation causing cerebrospinal fluid leak. The defect was patched by a piece of cartilage, and the surgical wound was closed with temporal muscle rotation flap and *blind sac closure*. The patient was discharged 5 days postoperative, and was given antituberculosis medication for 9 months. On 3<sup>rd</sup> week postoperative visit, the surgery wound was healed and she had normal blood sugar.

## CLINICAL QUESTION

“Could surgical management of malignant otitis externa with cranial nerve palsies complication in geriatric patients with multiple comorbidities achieve better result than conservative treatment?”

## REVIEW METHOD

Literature search using keywords ‘malignant otitis externa’ OR ‘temporal bone osteomyelitis’ AND ‘geriatric’ OR ‘elderly’ AND ‘multiple cranial nerve palsy’ AND ‘diabetes mellitus’ AND ‘tuberculosis’ AND ‘surgery’ OR ‘surgical’ was conducted through Cochrane, Pubmed, and Google Scholar.

## RESULT

From the search it was obtained 11 articles published in the last 10 years. Selection based on inclusion and exclusion criteria, found 2 studies were relevant with the topic.

DeSimone<sup>6</sup> reported a case of a 68 years old geriatric patient with chronic otitis externa caused by *Mycobacterium tuberculosis*. This was a very rare case. Several factors affecting the occurrence of tuberculosis otitis externa infection were the decreased of immune system and diabetes mellitus on elderly. Treatment of choice for TB otitis externa infection is extra pulmonal anti tuberculosis therapy (ATT) regimen for 12 months.

Rajput<sup>5</sup> described the cranial nerves involvement as a MOE complication. His study found 8 out of 21 MOE cases suffered cranial nerve palsy, in particular the facial palsy. However, neuropathies were not correlated well with the mortality.

Literature search did not find any *randomized controlled trial* article about the effectiveness of treatment. This was due to the fact that MOE caused by TB case was very rare.

## DISCUSSION

Aggressive infection occurs in MOE or osteomyelitis of temporal bone patients. In osteomyelitis, infection involves the bone's blood vessels, impairs the blood flow, and causes necrotic or infected bone, known as sequestra, as well as new bone formation around the area of necrosis. Osteomyelitis of the temporal bone is a rare disease that commonly occur secondary to otitis externa in immunocompromised patients, or to suppurative otitis media. Abrupt onset of symptoms and signs during the course of infection indicates an acute osteomyelitis.<sup>3,7</sup> In this patient, complication of cranial nerve palsy (CN.VII, IX, and X) was apparent that

it clinically supported MOE. The diagnosis was confirmed by MRI radiologic finding of wide destruction.

Based on the pathological dissemination, the temporal bone osteomyelitis can be divided into these three categories: osteomyelitis of the lateral temporal bone (bone involvement of the mastoid and middle ear cleft), osteomyelitis of the medial temporal bone (bone involvement of the otic capsule, petrous apex, and clivus), and pantemporal osteomyelitis (showing features of both mastoid-middle ear cleft and petrous involvement).

The osteomyelitis of temporal bone could originate from the surrounding area including chronic otitis media, chronic otitis externa, sphenoid sinusitis, or any inadequately treated chronic infection near the skull base. The infection may spread anteriorly involving the parotid gland, temporomandibular joint, or cranial nerve VII at the exit of the stylomastoid foramen. It may also spread posteriorly to the mastoid and vertical portion of cranial nerve VII, or inferomedially to the skull base including the carotid artery, the jugular bulb and the sigmoid sinus. Infection typically spreads through the haversian system of compact bone with progressive replacement of compact bone with granulation tissue. Bone destruction is both osteoplastic and osteoclastic.<sup>3</sup> Cranial nerves IX, X or XI palsies can occur when the jugular foramen becomes involved.

One of the most serious complications of MOE is the involvement of facial nerves. Facial nerve was found to be the most often affected, and this is due to the stylomastoid foramen is anatomically closer to the external auditory canal compared with the jugular foramen and is therefore more likely to be affected earlier. It might be the reason why facial nerve palsies did not resolved despite the infective disease process has been controlled, due to the fact that the anatomic length of nerve affected is longer compared with the other cranial nerves. It is also likely

that nerve involvement occurs because of infection or bacterial toxins directly inhibiting neurotransmission. Therefore, the longer the length of time that the nerve is exposed to toxins, the less likely it is to recover. As the facial nerve is usually the first nerve affected because of its proximity to the external auditory canal, it is liable to be affected for a longer period of time. In addition, because the patient was diabetic, blood supply to the facial nerve may be compromised by preexisting microvascular disease, and there may also be a degree of neuropathy affecting the nerve. Cranial nerve palsies do not affect MOE mortality.<sup>4,5</sup>

In this patient, the infection spread extensively to anterior, posterior, and inferomedial. The condition could be the result of decreased immune status of a geriatric patient with multiple comorbidities (diabetes mellitus, chronic kidney disease, and hypertension).<sup>3</sup> *Pseudomonas aeruginosa* was identified in this patient's swab culture of the external auditory canal. Most studies have shown that *Pseudomonas aeruginosa* is the most commonly isolated microbiological agent. However, in this patient an extensive caseous area was found during surgery, indicating a *Mycobacterium tuberculosis* infection, with secondary infection caused by *P.aeruginosa*. Tuberculosis manifestation in external auditory canal is very rare. Diagnosis of MOE due to TB is seldom considered in an immunocompetent adult host without pulmonary involvement. Often, patient did not show any pulmonary TB symptoms, therefore, temporal bone TB is very difficult to diagnose. Diagnosis of TB infection was built by the existence of caseous area, supported by a histopathologic examination. Bacterial culture test often cannot isolate acid-fast-bacillus, thus, any suspicion of TB should be supported by a histopathologic examination, because a routine culture of pus might not be able to isolate tubercle bacilli, and it also helps to diagnose early malignant change. If the diagnosis of extrapulmonary

tuberculosis was confirmed, a treatment of Anti Tuberculosis Therapy (ATT) must be administered, including isoniazid (300 mg daily), rifampicin (450mg), ethambutol (800mg), pyrazinamide (750 mg daily), for at least 9 months.<sup>3,6,8</sup>

Treatment of MOE consists of broad-spectrum antibiotics for not less than 3 months along with surgical debridement and a wide meatoplasty. Local treatment of the auditory canal includes meticulous cleaning and debridement. Use of topical antimicrobial agents is controversial because these drops will affect future culture results if there is no response to therapy. Once the diagnosis of skull base osteomyelitis has been confirmed, long-term antimicrobial therapy remains the mainstay of treatment with three general protocols: aminoglycoside and a  $\beta$ -lactamase antibiotic, a third generation cephalosporin (Ceftazidime), or an oral quinolone ciprofloxacin. However, there have been recent reports of resistance of *Pseudomonas* to ciprofloxacin therapy in temporal bone osteomyelitis. Treatment failures occur due to tissue hypoperfusion and hypoxia. In such cases the use of hyperbaric oxygen could increase wound partial pressure of oxygen levels, enhance phagocytic oxidative killing of aerobic microorganism, and promote angiogenesis and osteogenesis. Treatment consists of 100% oxygen given for 90 minutes at 2.5 atm. absolute pressure 5 days a week, 20 times as an adjuvant therapy.<sup>1,3,7</sup>

In the presence of severe pain, complications, refractory cases, or bony sequesters on High Resolution Computed Tomography (HRCT), surgical intervention is necessary. Prasad et al,<sup>3</sup> stated that surgery was the treatment of choice for most of the cases of chronic osteomyelitis due to the fact the presenting symptoms of severe pain and ear discharge, were seen in the majority of patients. Also, most of them had received prior antibiotic therapy that failed to resolve

the condition. Surgery treatment varied from wound debridement with skin preservation, mastoidectomy (canal up as well as canal down), to subtotal petrosectomy. The subtotal petrosectomy was done in cases of extensive granulations and sequestrations, including removal of all air cells of the temporal bone leaving behind the otic capsule and the petrous apex. The external auditory canal was closed as a blind sac and the cavity obliterated with temporalis muscle flap. Temporal bone resection and petrosectomy have surgical risks such as cerebrospinal fluid leak, hematoma, wound infection, and death. The mortality rate of MOE is 10-20%.<sup>3,7</sup>

The decision to operate this patient had to consider the comorbidities and her geriatric factors. A multidisciplinary assessment was implemented by a team of specialists consisted of Nephrologist, Metabolic-Endocrinologist, Geriatrician, Otorhinolaryngologist, Clinical Nutritionist, Medical Rehabilitation, and Critical Care Physician (Intensivist).

Comprehensive preoperative assessment to ensure patient's safety should be taken prior to surgery. Patient had anemia and hypoalbuminemia which could probably influence the surgery success rate particularly in wound healing and prevention of infection aspects. The clinical nutritionist should plan the optimal nutrition intake. As to ensure adequate intake route, if necessary, nasogastric tube could be inserted.

The perioperative blood sugar level should be intensively monitored. Patients' blood sugar level was under control up till 3 weeks postoperative. Afterwards, patient went back to her hometown. Perioperative control of blood sugar by anesthesiologist helped to minimize the infection risk of the surgical wound and raised the recovery chance of sepsis patients. The postoperative blood sugar control better be performed with intravenous insulin injection or intravenous drip rather than subcutaneously. The capillary blood sugar level should be maintained under 200 ml/dl in 48 hours postoperative.<sup>3,7,9</sup>

Medical rehabilitation had an important role in management of cranial palsies. For unresolved postoperative peripheral facial nerve palsy, should be considered to rehabilitate facial nerve function by relaxation, massages, and facial muscle exercise.<sup>4,5</sup> Dysphagia was also present in this patient, which need rehabilitation of swallowing function.

Temporal bone osteomyelitis is a rare case. An Otolaryngologist should think of it when finding patient with complaints of otalgia accompanied by a history of diabetes mellitus, particularly in patients who do not response to standard therapy. Supporting examination tests should be conducted, such as bacterial culture and histopathologic examination to find atypical causes such as tuberculosis infection. Long-term antibiotics medication is still a therapy-of-choice for MOE. However, in an aggressive osteomyelitis, operative intervention must be performed. High complexity patients require a multidiscipline teamwork to discuss and decide the optimal treatment.

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