

Case Report

Osteoradionecrosis and cholesteatoma of external auditory canal in post-radiotherapy nasopharyngeal carcinoma patient

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ABSTRACT

Background: Osteoradionecrosis and cholesteatoma of the external auditory canal following external-beam radiotherapy as the treatment of nasopharyngeal carcinoma is a rarely found complication. Patients with external auditory canal cholesteatoma (EACC) typically present with chronic otorrhea and dull pain due to the local invasion of squamous tissue into the bony external auditory canal (EAC). **Purpose:** To remind ENT specialists and general practitioners about the risk osteoradionecrosis and cholesteatoma of external auditory canal in nasopharyngeal carcinoma patient after radiotherapy treatment. **Case:** We report a case of osteoradionecrosis and cholesteatoma of EAC in nasopharyngeal carcinoma (NPC) patient with complaint of a foul-smelling discharge from her right and left ears. Two years previously she had undergone external-beam radiotherapy to the neck as the treatment for nasopharyngeal carcinoma. **Management:** The cholesteatoma was removed microscopically on local anesthesia. After the cholesteatoma had been removed the right ear result of pure tone audiometry showed mild degree conductive hearing loss (27,7 dB), while the left ear within normal hearing threshold. **Conclusion:** Osteoradionecrosis and cholesteatoma of external auditory canal could develop as a complication of radiotherapy in nasopharyngeal carcinoma patient.

Keywords: osteoradionecrosis, cholesteatoma, radiotherapy, chronic otorrhea.

ABSTRAK

Latar belakang: Komplikasi osteoradionekrosis dan kolesteatoma pada liang telinga luar akibat radioterapi pada pengobatan karsinoma nasofaring sangat jarang terjadi. Pasien dengan kolesteatoma liang telinga luar biasanya datang dengan keluhan otore kronis dan nyeri akibat invasi dari jaringan skuamus ke tulang liang telinga luar. **Tujuan:** Memberi wawasan bagi dokter umum dan spesialis THT-KL tentang adanya risiko osteoradionekrosis dan kolesteatoma pada liang telinga luar akibat radioterapi pada penderita karsinoma nasofaring. **Kasus:** Dilaporkan satu kasus osteoradionekrosis dan kolesteatoma di liang telinga luar pada penderita karsinoma nasofaring dengan keluhan sekret telinga berbau busuk pada liang telinga kanan dan kiri. Dua tahun sebelumnya pasien tersebut mendapat radioterapi untuk pengobatan karsinoma nasofaring. **Penatalaksanaan:** Kolesteatoma diangkat secara mikroskopis dengan anestesi lokal. Pemeriksaan audiometri nada murni pascatindakan didapati tuli konduktif derajat ringan (27,7dB) pada telinga kanan sedangkan telinga kiri dalam batas normal. **Kesimpulan:** Radionekrosis dan kolesteatoma liang telinga luar merupakan komplikasi terapi radiasi pada kasus karsinoma nasofaring.

Kata kunci: osteoradionekrosis, kolesteatom, radioterapi, otore kronis.

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INTRODUCTION

Osteoradionecrosis (ORN) is one of the most serious complication caused by radio-

therapy treatment in nasopharyngeal carcinoma (NPC) patients. ORN rarely occurs in the external auditory canal. Radiotherapy is

the mainstay treatment of nasopharyngeal carcinoma (NPC) with the classic 3 H principle of Hypoxia, Hypovascularity and Hypocellularity, but it may actually impairs normal collagen synthesis and cell production, leading to tissue breakdown and eventual radionecrosis.¹ ORN, though not commonly seen nowadays, is one of the most serious complications of radiotherapy in NPC.² It may occur in the skull base,^{1,2} mandible,^{1,3} maxilla and external auditory canal in NPC patients. Signs of ORN include unhealed ulcer, exposed bone and accompanying granuloma, all of which closely mimic signs of malignancy and pose a significant challenge for differentiation.^{1,3}

External auditory canal ORN is seldomly reported in the literature.^{1,4-7} It is probably overlooked by many clinicians because the initial symptom of crust, otorrhea, and otalgia may lead to the diagnosis of chronic otitis media instead. However, potentially disastrous complications, such as malignant otitis externa, meningitis or brain abscess, may occur and resulted in death.⁷⁻¹²

We report a nasopharyngeal carcinoma case with osteoradionecrosis and cholesteatoma of the external auditory canal after external-beam radiotherapy treatment in a woman who had experienced this complication 2 years after she had undergone 32nd times radiotherapy. Her condition resolved after removal of crust from the external auditory canal, followed by antibiotic therapy and periodic aural toilet with local anesthesia.

CASE REPORT

A 39-year-old woman complained of a foul-smelling discharge from her right and left ears since 4 months previously and she had used ear drop antibiotics but got no improvement. Two years before she had undergone external-beam radiotherapy to the neck as the treatment for nasopharyngeal carcinoma.

Otoscopic examination of the right ear showed hyperaemic with crust of the external auditory canal. The left ear was hyperaemic and the external auditory canal was filled with crust, which after removal of the crust, the tympanic membrane was seen intact. The external auditory canal was evaluated and found that the posterior part of the skin was destroyed until the bone was exposed. Histologic examination of the aural crust revealed keratinizing squamous cell of the epithelium (cholesteatoma).

After the cholesteatoma had been removed, a strip of gauze with topical antibiotics was deposited in external ear canal for three days and after the removal of the gauze, she got topical antibiotic treatment and regular aural toilet.

As for the right ear, the result of pure tone audiometry showed mild degree conductive hearing loss (27,7 db), and both of the tympanometry was normal, while the left ear within normal hearing threshold.

Computed tomography of the temporal bone did not suggest middle ear pathology, no erosion in the temporal bone that was compatible with osteoradionecrosis and showed an EACC as a soft-tissue mass in the inferior EAC, with associated erosion of the subjacent bone.

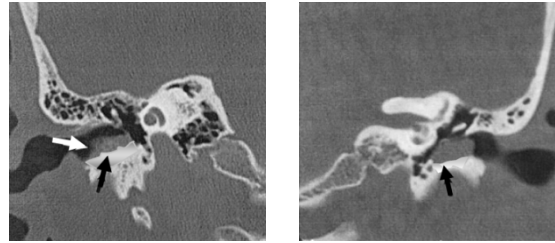


Figure 1: Coronal temporal bone CT image shows an EACC as a soft-tissue mass in the inferior EAC, with associated erosion of the subadjacent bone.

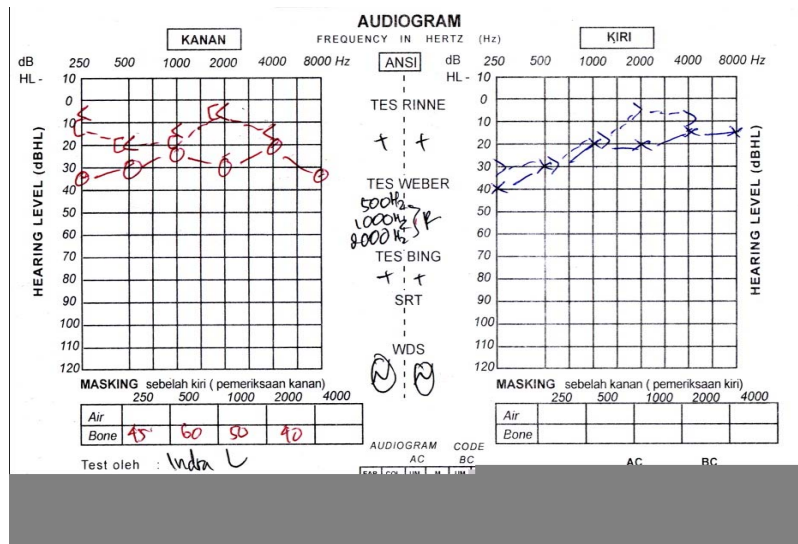
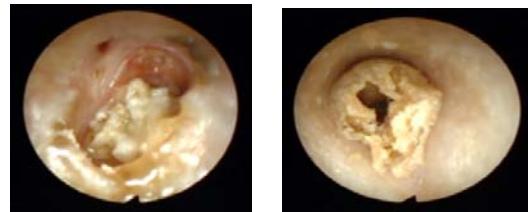


Figure 3: Pure tone audiometry

DISCUSSION

Osteoradionecrosis and cholesteatoma of the external auditory canal following external-beam radiotherapy as the treatment of nasopharyngeal carcinoma is a rarely found complication following a long post treatment interval, in this case 2 years. Patients usually present with chronic, offensive otorrhea and occasionally otalgia.^{1-4,12}

External auditory canal cholesteatoma (EACC) is a rare entity with an estimated occurrence of one in 1000 new patients at otolaryngology clinics. Patients with EACC typically present with chronic otorrhea and dull pain due to the local invasion of squamous tissue into the bony EAC. Most cases are spontaneous or occur after trauma to the auditory canal, though pre-existing

ear-canal stenosis or obstruction has also been reported to produce EACC. The clinical differential diagnosis including neoplasms of the EAC and inflammatory or infective conditions such as keratosis obturans, postinflammatory medial canal fibrosis, and malignant otitis externa.^{7,8,12}

A cholesteatoma is a cystic structure lined by keratinizing stratified squamous epithelium with associated periostitis and bone erosion, which is most commonly found in the middle ear cavity. Middle ear cholesteatomas may be congenital or acquired, but approximately 98% are acquired. Although cholesteatomas are found almost exclusively in the middle ear and mastoid, in rare cases they occur in the EAC. The estimated incidence of EACC is 0.1–0.5% of all otologic patients. Although EACC is rare, recognizing it as a distinct entity is important because its management is notably different from that of its clinical differential diagnoses.^{7,9,10}

The exact etiology of EACC is unclear. Most cases are spontaneous or occur after trauma in the auditory canal, although ear canal stenosis or obstruction has also been described as a causative factor. Trauma, either canal skin lacerations, may isolate the squamous epithelium or cause stenosis of the canal; either of these events could lead to EACC.^{1,7-11}

Normal epithelial migration from the tympanic membrane and EAC is an important self-cleansing property of the outer ear. Epithelial migration carries the keratin

debris laterally outward from the tympanic membrane for removal. Spontaneous EACC has been described as a disease of the elderly due to the loss of normal migration in aging epithelium of the canal wall. Another pathophysiologic hypothesis suggests that accumulation of keratin debris induces changes of cellular proliferation in the EAC.^{1,7}

Other literatures described a lower than average migratory rate of epithelium in the inferior wall of the ear canal in cases of EACC. The cause of EACC may be partly related to abnormal epithelial migration, which leads to the local accumulation of squamous epithelium that can evolve into an EACC. In addition, the decreased migratory rate is thought to be related to a poor blood supply.^{1,7}

Imaging can be valuable in evaluation of EACC. In temporal bone CT examination, EACC is most commonly seen as an EAC soft-tissue mass with associated bone erosion and intramural bone fragments. The bone erosion adjacent to the soft-tissue mass may be smooth, similar to a middle ear cholesteatoma; however, the erosion may be irregular secondary to the necrotic bone and periostitis.^{7,10}

Patients with EACC usually present with chronic otorrhea, dull pain and less commonly, they present with hearing loss. Gross pathologic analysis of EACC demonstrates extensive erosion of the bony EAC by a wide mouthed stratified squamous keratini-

zing epithelial sac with a localized periostitis and sequestration of bone. The tympanic membrane is typically normal. The interface between the EACC and the bone is erosive. This erosion is thought to be related to proteolytic enzymes along the margin of the lesion produced within the cyst lining; these weaken the bone and result in periostitis and sequestration of bone. The erosion could also be partly related to the accumulation of keratin debris, which traps moisture and results in a bacterial infection that can cause ulceration of the epithelial layer and granulation tissue formation in patients who have a superimposed infection.⁷

In the diffuse type, a widespread ischemic osteonecrosis involves the skull base and adjacent structures. These patients have usually received higher doses of external irradiation to the temporal bone. Severe otalgia and pulsatile, offensive otorrhea are common. Cranial nerve palsies might also be present. Diffuse osteoradionecrosis is associated with a recognized incidence of local or regional complications, such as suppurative labyrinthitis, trismus, meningitis, cerebrospinal fluid leakage, and internal carotid aneurysm.³⁻⁶

On otoscopic examination, EACC can be difficult to distinguish from other inflammatory, infective, or neoplastic processes of the EAC; examples of these include keratosis obturans, postinflammatory medial canal fibrosis, malignant otitis externa, and squamous cell carcinoma (SCC). Keratosis

obturans is the most closely related condition and the one most difficult to distinguish. In fact, keratosis obturans was previously considered to represent the same disease process as EACC.⁷

The diagnosis is established on the basis of the history and clinical examination and audiometric findings. Imaging is usually not performed for diagnosis. Surgical management is the treatment of choice. Interestingly, EACC has been described in association with postinflammatory medial canal fibrosis in rare cases.^{7,9,10}

The management of osteoradionecrosis in the external auditory canal by conservative treatment with frequent aural toilet and topical antibiotics is often administered for localized osteoradionecrosis.^{4,6,12}

Radiation exerts the 3 Hs of hypoxia, hypovascularity and hypocellularity, impairs normal collagen synthesis and cell production, and leads to tissue breakdown and eventual ORN. ORN of the external auditory canal typically occurs in the lower portion of the canal, which can be explained by the downward pressure exerted by wearing a hearing aid or picking at ear wax. The pressure may also involve the perichondrium that carries the main blood supply to the cartilage, resulting in pressure necrosis. Then, microorganisms may penetrate through the pressure ulcer, leading to serious perichondritis and chondritis.^{1,4-9}

The principle of treating ORN is basically surgical, as the necrotic substance

cannot be revitalized and has to be surgically removed.³ The lack of vascularized tissue around the ORN site further complicates treatment. Prevention of infection with antibiotics and maintaining local hygiene by frequent cleansing are also mandatory in controlling the disease.^{3,4,8}

The symptomatology of external auditory canal ORN of purulent otorrhea, otalgia, exposed necrotic bone and granuloma, may mimic that of chronic otitis media, external auditory canal cholesteatoma or ear malignancy. The differentiation between ORN and recurrent cancer might be difficult.^{2,5-9}

NPC patients with external auditory canal ORN typically present with foul odor because of the sequestrum and thus may be socially repelled. The symptoms of otorrhea or otalgia, which mimic those of chronic otitis media, may be neglected by patients. The disease is also ignored by clinicians, probably because of unawareness of the disease entity.^{2,4-8}

ORN involving the external auditory canal is quite unusual in NPC patients. The clinical presentations of otorrhea, otalgia and crust formation are very similar to those of chronic otitis media, external auditory canal cholesteatoma or malignancy. A high suspicion index is mandatory for early diagnosis. The disease may lead to disastrous complications and should never be neglected by clinicians. CT evaluation could help the diagnosis by showing soft-tissue attenuating mass in the EAC with erosion of adjacent

bone, and bone fragments are often present within the mass. The management should be by surgical evacuation of the pathological entities.

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