A Rare Progression of Malignant Otitis Externa: A Case Report

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Abstract
Malignant otitis externa (MOE), although not a malignancy by itself, is a serious life threatening necrotizing infection of the external auditory canal and lateral skull base. Toulmouche is the pioneer in describing this condition.¹ The first case of MOE was reported in 1938, and the term malignant otitis externa was coined later by Chandler in 1968 due to its high fatality rate. The incidence of MOE is higher in immunocompromised individuals like uncontrolled diabetics (86-90%), HIV and in patients on long term immunosuppressive therapy for various systemic conditions. Most common pathogen involved is Pseudomonas aeruginosa(99.2%).³ MOE can spread to involve the adjacent structures resulting in skull base osteomyelitis which has different routes of spread. We report a rare case of malignant otitis externa in an elderly diabetic male with mandibular condyle osteomyelitis with an unusual anterior spread and caused by a relatively rare pathogen.

Keywords: Malignant otitis externa, uncontrolled diabetes, mandibular condyle osteomyelitis, anterior spread

Introduction

Case Presentation:
A 70 years old male, known diabetic, under control since 10 years came to the department of ENT with chief complaints of on and off left sided ear pain aggravated at night since 3 months. There was also a h/o ear discharge which was purulent since 1 month. No h/o recent trauma. H/O hearing loss along with pain on opening his mouth and chewing was present.

On clinical examination, there was swelling of the left pre auricular region extending down upto the angle of mandible. (Figure 1) The swelling was diffuse, soft and tender on palpation with the overlying skin being normal. No evidence of any discharging sinus. Otoscopic examination revealed a dense aural granulation at the osseous cartilaginous junction which was pulsatile in nature. (Figure 2) Pure tone audiometry showed mixed hearing loss. Routine blood investigations revealed neutrophilic leukocytosis and raised ESR – suggesting an active underlying infection.

A sterile swab from the infected left ear was taken concurrently and sent for gram staining along with culture and sensitivity. An aural toileting was given subsequently.

Later, on admission, patient was referred for high resolution computed tomography of temporal bone which showed soft tissue thickening of the left external auditory canal at the osseocartilaginous junction with extensive erosion of the anterior wall of bony external auditory canal leading to mandibular condyle osteomyelitis. There was also a soft tissue density occupying the entire left masticator space. (Figure 3, 4a) MRI showed evidence of soft tissue mass with adjacent edema and effacement of fat planes involving the left masticator space and infiltration of bone marrow. (Figure 4b). A diagnosis
of malignant otitis externa causing mandibular condyle osteomyelitis was made on clinical and radiological grounds.

The patient was treated empirically with oral ciprofloxacin along with regular aural toileting and other conservative measures. Patient’s diabetes status was under check with oral hypo glycemic drugs in order to reap the benefits of anti-microbials.

Later, the swab turned out to be positive for coagulase negative Staphylococcus aureus, sensitive to ciprofloxacin and hence was continued with the same management for a period of 4 weeks. As a microbiologic diagnosis is crucial for correct treatment, a biopsy from the granulation tissue was taken which revealed non specific inflammatory cells with no substantive evidence of growth, thereby ruling out malignancy and untangled the diagnosis of necrotizing otitis externa.

Discussion:

Malignant otitis externa, also known as necrotizing otitis externa is an infection of the external ear canal usually the osteo-cartilaginous junction with involvement of skull base resulting in skull base osteomyelitis(SBO). Mortality rates ranges from 28 to 60% along with cranial nerve palsy in cases of delayed diagnosis and treatment.

Endarteritis and microangiopathy leading to hypoperfusion and immune dysfunction in diabetics plays an important role in the pathogenesis of this condition, however there are also cases reported in non diabetics. Most common pathogen isolated in diabetics was Pseudomonas aerogenosa(99.2%). Fungal infections have also been reported in 5% to 20% of the cases, most common being Aspergillus fumigatus. Other rare pathogens isolated are Proteus, Staphylococcus and Klebsiella. Our case is one such rare entity where Staphylococcus was the etiologic agent. Withal, unlike typical cases of MOE which involves the TMJ, panoramic images in this case showed cortical erosion of left mandibular condyle, as literature evidence shows only 18 such cases.

Clinical features of MOE include severe otalgia, purulent otorrhea and hearing loss. Temporomandibular joint pain, hemifacial pain, headache, and trismus can develop from the anterior extension of the disease. The infection can spread anteriorly to the masticator space, condylar bone marrow and temporomandibular joint, medially to the pharyngeal mucosal space to involve the vagus and glossopharyngeal nerve. It can show extensive intracranial involvement as well to reach the sigmoid sinus, jugular vein and ICA, posteriorly to the mastoid process and stylomastoid foramen via the fissure of Santorini and hence can involve the facial nerve as well.

Although CT and MRI helps in detecting bony erosions and extent of soft tissue involvement respectively, Radio isotope scans (Gallium citrate 67/Tc 99) are useful in monitoring the resolution of the disease in response to treatment, reflected by the intensity of uptake.

Management is usually multidisciplinary, spreading over strict glycemic control, antibiotic mediated therapy and surgical debridement. Literature shows long term antibiotic therapy yields the same results, even without surgical procedures as evident in our case. Surgical treatment is reserved for cases where an abscess has formed in the joint space. Our patient showed improvement with antibiotics and other supportive measures as evident by resolution of the pre auricular swelling and the aural granulation(Figure 5), hence not requiring any invasive procedures.

Conclusion:

Diagnosis of condylar osteomyelitis in the setting of MOE caused by Staphylococcus aureus in a controlled diabetic status is extremely rare and is frequently delayed because of its nonspecific symptoms, long clinical course, and radiologic findings that usually mimic those of malignancy. Therefore, it requires a global perspective of the patients who are at risk, recognition of the more typical radiologic features, and inclusion of this rare entity in the differential diagnosis for a refractory otitis externa. An understanding of the anatomical routes of spread of infection, unusual causative pathogen is essential to make an early diagnosis to reduce the mortality rates due to this serious condition, thereby improving the clinical outcome.

References:

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2. Temporomandibular joint disorder from skull-base osteomyelitis: a case report Suck-Chul Lee, Jae-Hyung Kim, Chul-Hoon Kim and Bok-Joo Kim*


Figure 1: Pre auricular swelling on the left side with no evidence of discharging sinuses.

Figure 2: Otoscopic examination revealed a pale pink aural granulation tissue at the osteo cartilaginous junction.
Figure 3: Axial non contrast CT showing a homogenous soft tissue density occupying the entire left masticator space.

Figure 4a: NCCT of the skull base, coronal reconstructed bone window showing bony erosions of the left mandibular condyle. Preserved clivus

Figure 4b: T2W MRI, axial section showing soft tissue mass with adjacent edema and effacement of fat planes involving the left masticator space
Figure 5: Post 4 weeks of antibiotics. Otoscopy (5a) showing marked reduction in size of granulation and 5b showing resolution of the pre auricular swelling clinically.