A Unique Case of Malignant Otitis Externa

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A 11-year-old male presented to the hospital with a 2-day history of right otalgia, purulent otorrhea, and swelling after hot tub exposure. There was no significant medical, family, or social history. On physical examination, the right ear canal contained purulence with significant erythema, edema, and tenderness along the lobule, tragus, and preauricular skin (Figure 1). The rest of the head and neck examination, including the left ear, was normal.

A computed tomography scan showed a soft tissue phlegmon, subcutaneous emphysema, gas along the pinna, edema around the mastoid tip and skull base, and bony changes suggestive of malignant otitis externa (MOE; Figure 2). After consultation with the infectious disease service, intravenous (IV) meropenem was started due to concern for bony involvement that was continued for 1 day. He improved and was discharged on day 3 but returned 1 week later with increasing pain and drainage. A combination of IV meropenem, clindamycin, and amphotericin was started by the infectious disease service. He was discharged on day 4, after which he underwent four serial debridements of the tragus, concha, and lateral external auditory canal (EAC) in the operating room over 21 days, with repeated cultures growing Actinomyces. A 6-month course of oral doxycycline was initiated, with local wound care in clinic every 2 weeks, along with ciprofloxacin–dexamethasone ear drops twice daily. A keloid of the right lobule developed at month 4, which was treated with Kenalog steroid injections at months 5 and 7. At 8-month follow-up, there was no evidence of MOE or EAC stenosis (Figure 3), and audiometric testing confirmed normal hearing and tympanic membrane function. Final workup for autoimmune disorders, immunodeficiencies, and diabetes was negative.

We present a case of culture confirmed Actinomyces MOE in an immunocompetent child. Otitis externa can involve the superficial ear up to the tympanic membrane, while MOE comprises an aggressive infection of the EAC that spreads into the mastoid and skull base, with or without bony erosion.1,2 Classically, MOE is seen in patients with diabetes, immunocompromise, or malignancy.3 However, there have been selective cases of MOE in immunocompetent adults.4,5

Figure 1. Initial presentation showing purulent right external auditory canal tissue.

aeruginosa is causative in over 90% of cases, but Staphylococci, Streptococci, and gram-negative bacteria have also been implicated.2,6

Recent studies on MOE have reported increased pathogen diversity. In addition to increased rates of quinolone-resistant Pseudomonas, there have been reported cases of Streptococci, Staphylococci, and gram-negative pathogens.2,7 Fungal involvement has been implicated in patients with immunosuppression or uncontrolled diabetes.8 There has been an increase in culture-negative cases, most likely due to overuse of antipseudomonal antibiotics.9 Thus, although Pseudomonas

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predominates in cases with MOE, other organisms should be considered.

Actinomyces is a rare gram-positive, anaerobic bacterium that enters tissue via disruptions in mucosal surfaces and typically causes chronic, granulomatous infections. In the head and neck, Actinomyces infections usually occur in the oral–cervicofacial area, particularly oral and dental infections.10 Ear infections tend to present with chronic mild otorrhea or otitis media.11 Treatment has involved at least 1 surgical debridement and an extended course of oral antibiotics for up to 6 months, with nearly all cases reporting complete resolution without complication. To our knowledge, there are no previously reported cases of Actinomyces-implicated MOE.

Regular debridement in cases of MOE is rare, even in cases requiring hospitalization.12 The use of debridement has decreased with the advent of antipseudomonal antibiotics. However, with increasing rates of quinolone-resistant Pseudomonas and non-Pseudomonas pathogens, prompt debridement may be warranted if there is no response to initial antibiotic treatment. Cases of MOE in nondiabetic or immunocompetent patients, although rare, may also represent infection with particularly resistant pathogens. A lower threshold for early and serial debridements may be required in these cases to provide histologic samples and therapeutic benefits.

Although rare in children, MOE should remain in the differential for any child presenting with severe otalgia, purulent drainage, and significant inflammatory signs, given the potential significant complications. Prompt antibiotic therapy remains the standard of care, but early serial debridements should be considered, given the increased microbial diversity recently implicated. Serial surgical debridements can reduce the burden of necrotic tissue and provide tissue samples for accurate microbial diagnosis.

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