

Malignant Otitis Externa Caused by Atypical Pathogens: A Report of Two Cases with Atypical Clinical Courses

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ABSTRACT

Malignant otitis externa is an aggressive infectious process of the external auditory canal, which may progress to involve adjacent structures including the skull base. It typically occurs in diabetics or patients with other underlying immunodeficiency, and is most commonly due to *Pseudomonas aeruginosa*. This poster reports two cases of malignant otitis externa caused by atypical pathogens. Both patients had underlying immunosuppression and were initially managed with oral and intravenous antibiotics and serial office debridements. Both patients were poorly compliant with treatment regimens, leading to progressive disease and the need for operative intervention. Specimens obtained from operative debridement and mastoidectomy eventually grew *Dermabacter hominis* in one patient and *Finegoldia magna* in the other. These organisms rarely cause infectious pathology in adults, and have not yet been described as causative pathogens in cases of chronic malignant otitis externa. The identification of these pathogens underscores the need for culture-directed therapy in patients being treated for malignant otitis externa.

REFERENCES

- Chen CN, Chen YS, Yeh TH, Hsu CJ, Tseng FY. Outcomes of malignant external otitis: survival vs mortality. *Acta Otolaryngol* 2010; 130:89-94.
- Hobson CE, Moy JD, Byers KE, Raz Y, Hirsch BE, McCall AA. Malignant Otitis Externa: Evolving Pathogens and Implications for Diagnosis and Treatment. *Otolaryngol Head Neck Surg* 2014; 151:112-116.
- Fernández-Natal I, Sáez-Nieto JA, Medina-Pascual MJ, Albersmeier A, Valdezate S, Guerra-Laso JM, Rodríguez H, Marrodán T, Parras T, Tauch A, Soriano F. *Dermabacter hominis*: a usually daptomycin-resistant gram-positive organism infrequently isolated from human clinical samples. *New Microbes New Infect* 2013; 1:35-40.
- Rosenthal ME, Rojtman AD, Frank E. *Finegoldia magna* (formerly *Peptostreptococcus magnus*): an overlooked etiology for toxic shock syndrome? *Med Hypotheses* 2012; 79:138-140.
- de Moreuil C, Héry-Arnaud G, David CH, Provost B, Mondine P, Alavi Z, de Saint Martin L, Bezon E, Berre RL. *Finegoldia magna*, not a well-known infectious agent of bacteriemic post-sternotomy mediastinitis. *Anaerobe* 2015; 32:32-33.
- Martin J, Bemer P, Touchais S, Asseray N, Corvec S. Recurrent abscesses due to *Finegoldia magna*, *Dermabacter hominis* and *Staphylococcus aureus* in an immunocompetent patient. *Clinical Microbiol* 2009; 15:201-203.
- Berenholz L, Katzenell U, Harell M. Evolving resistant *Pseudomonas* to ciprofloxacin in malignant otitis externa. *Laryngoscope* 2002; 112:1619-1622.
- Amorosa L, Modugno GC, Pirodda A. Malignant external otitis: review and personal experience. *Acta Otolaryngol Suppl* 1996; 521:3-16.

CASE 1

A 58-year-old male with a history of type 2 diabetes mellitus, coronary artery disease, congestive heart failure, and hepatitis B was referred to our otology clinic with bilateral otorrhea, otalgia, and hearing loss refractory to courses of topical Ciprodex. Physical examination and CT scan of the temporal bones revealed erosion and soft tissue opacification of the bilateral external auditory canals, with sparing of the middle ear and mastoid. He was started on oral ciprofloxacin and topical Ciprodex, and underwent serial office debridements. Cultures obtained in the office repeatedly grew *Corynebacterium* species. He failed to improve, and subsequently was started on IV vancomycin, which was transitioned to PO linezolid due to elevated transaminases. Severe thrombocytopenia occurred and linezolid was discontinued. He developed worsening disease that now extended to the middle ear and mastoid, with severe tympanic membrane retraction and cholesteatoma. He underwent left canal wall down mastoidectomy and right ear debridement, with intra-operative cultures revealing *Dermabacter hominis*. Post-operatively his left ear was noted to be healing well. He had no further left-sided otorrhea and reported improvement in his hearing. Given his positive response to surgery, right-sided mastoidectomy was planned, but in the interim he was admitted to the medicine service for severe sepsis secondary to *Pseudomonas* bacteremia. The source was felt to be from either a pneumonia or urinary tract infection. Otologic examination upon admission did not reveal any active otorrhea or purulence in either ear. He rapidly declined during his hospitalization, suffering a cerebrovascular accident and developing respiratory failure, and he ultimately expired.

CASE 2

A 56-year-old female with alcoholism was referred for bilateral otorrhea, otalgia, and hearing loss refractory to treatment with oral and topical antibiotics. Physical examination revealed necrotic bone with purulent otorrhea in the bilateral external auditory canals. She was started on topical Cortisporin but was poorly compliant. Operative intervention was planned. Pre-operative imaging revealed aggressive external otitis and mastoiditis with bony erosion of the external canals, mastoids, and temporomandibular joints, as well as a 2.5cm parapharyngeal space abscess. She underwent incision and drainage of the left parapharyngeal space abscess and bilateral debridement of the necrotic bone in the operating room. Cultures revealed *Finegoldia magna* from the ear canals, and gram-positive cocci in the parapharyngeal space. She left the hospital against medical advice on post-operative day two. She was seen in clinic once after discharge, at which time she continued to be poorly compliant with her antibiotic therapy. She has not attended any additional follow-up appointments and has been lost to follow-up.

DISCUSSION

Malignant otitis externa is an aggressive infectious process of the external auditory canal and mastoid, which may progress to involve adjacent structures including the skull base. It typically occurs in patients with underlying immunodeficiency. *Pseudomonas aeruginosa* is the most common causative organism; however, recent studies have shown that *Pseudomonas* infection may be less common than previously believed.¹ Alternative pathogens, such as methicillin-resistant *Staphylococcus aureus*, have been increasingly implicated.² In these case reports, we describe two cases caused by atypical pathogens.

Both patients described in this report had underlying immunosuppression, one from diabetes mellitus and hepatitis B, and the other from alcoholism. Both patients presented with similar complaints and were managed medically with topical and oral antibiotics and in-office debridements. While each patient initially followed a fairly indolent clinical course, both patients eventually required operative intervention for progressive disease.

In the first case, intra-operative cultures grew *Dermabacter hominis*, a gram-positive, rod-shaped, lactose-fermenting pathogen which was formerly identified as a *Coryneform* bacteria. While this organism rarely causes infectious pathology in adults, it has infrequently been implicated in cases of acute otitis externa.³ Diagnosis of this organism may be challenging as it is commonly confused with *Corynebacterium* species. Interestingly, this patient responded well to unilateral tympanomastoidectomy with both subjective and objective improvement in the operative ear. He was offered contralateral tympanomastoidectomy, but died from sepsis secondary to a *Pseudomonas* bacteremia prior to the planned operation. There was no culture data or physical exam findings to suggest an otologic source of his sepsis.

In the second case, intra-operative cultures grew *Finegoldia magna*. *Finegoldia magna* is an anaerobic gram-positive coccus which was previously classified as *Peptostreptococcus magnus*. It is normal flora of the skin and oral cavity, but has virulent potential.^{4,5} It may also develop resistance to antimicrobials targeted at anaerobic bacteria, including clindamycin.⁶

To our knowledge, these are the first two cases suggesting these pathogens as causative agents in cases of malignant otitis externa. The presence of these atypical pathogens underscores the changing epidemiology of the disease and the importance of obtaining reliable culture data from which to direct treatment. Oral and topical fluoroquinolone therapy has traditionally been the mainstay of therapy in order to empirically treat for *Pseudomonas* infection. However, fluoroquinolones have poor gram-positive coverage, and these organisms are increasingly being identified as causative organisms in cases of malignant otitis externa. In addition, some *Pseudomonas* strains may be resistant to fluoroquinolones.⁷ If patients do not respond to initial treatment with fluoroquinolones, cultures should be obtained and practitioners should consider broadening antibiotics to cover gram-positive organisms, including MRSA.

This disease is not typically managed surgically. In rare cases, surgery may be offered and includes radical mastoidectomy with partial resection or debridement of the skull base and infratemporal fossa, and often microvascular free tissue transfer reconstruction. Previous case series have shown that radical debridement did not arrest disease progression.¹ Other authors have suggested that surgical management may only serve to expose healthy bone to the pathogen, leading to worsening disease.⁸ Our experience in Case 1 differs from these reports. Our patient was offered canal wall down tympanomastoidectomy. The patient responded well to surgery; he had no further otorrhea and reported a subjective improvement in his hearing. This suggests a potential role for surgical therapy earlier in the disease course than traditionally understood.

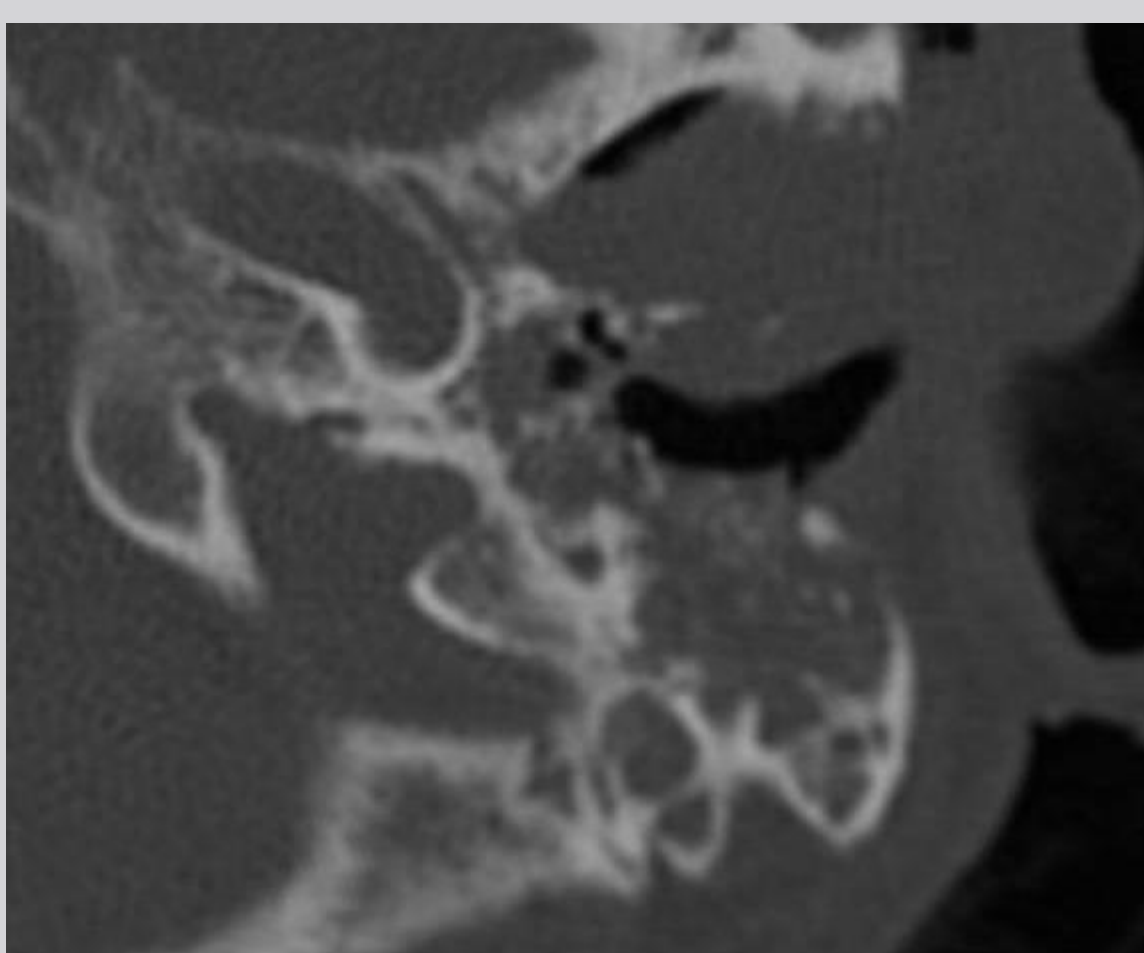


Figure 1. Pre-operative axial CT of left temporal bone of the patient in Case 1 showing progression of disease, with bony erosion of the external auditory canal, mastoid cavity, and middle ear.



Figure 2. Intra-operative photo of the patient in Case 1 showing extensive necrotic bone which was debrided

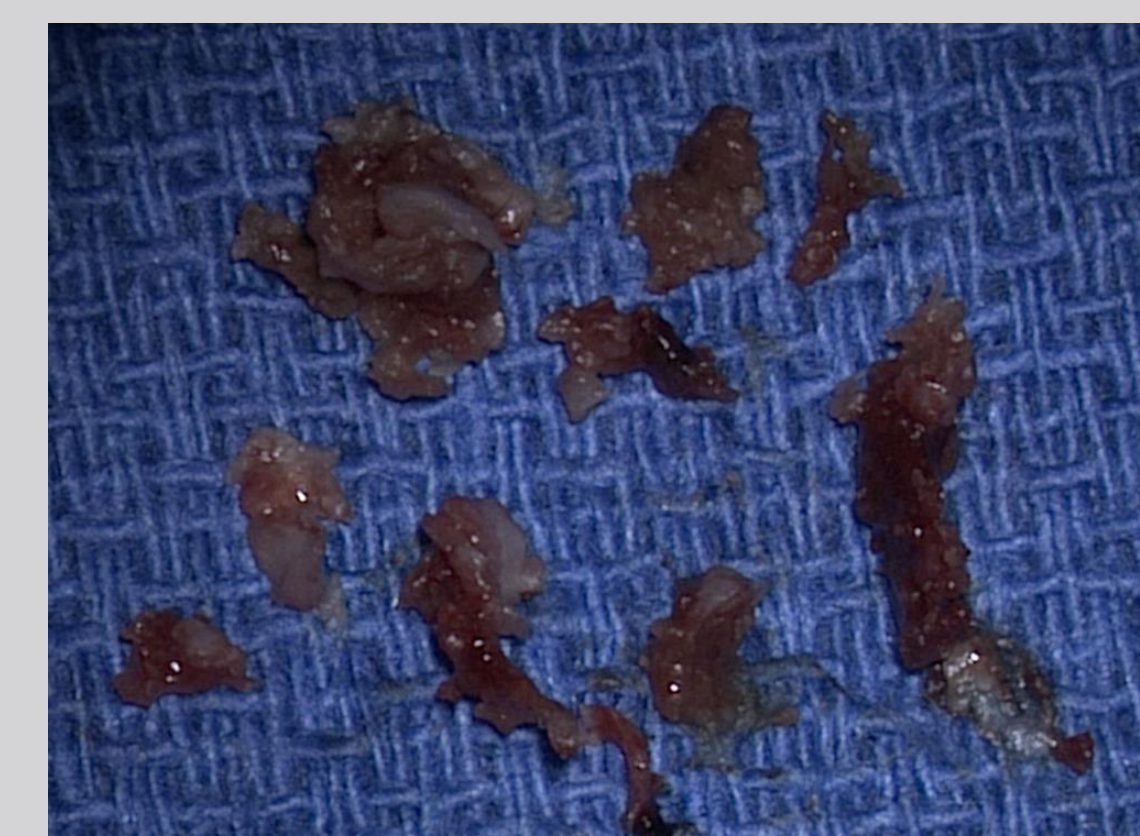


Figure 3. Necrotic bone debrided from the external auditory canal of the patient in Case 1.

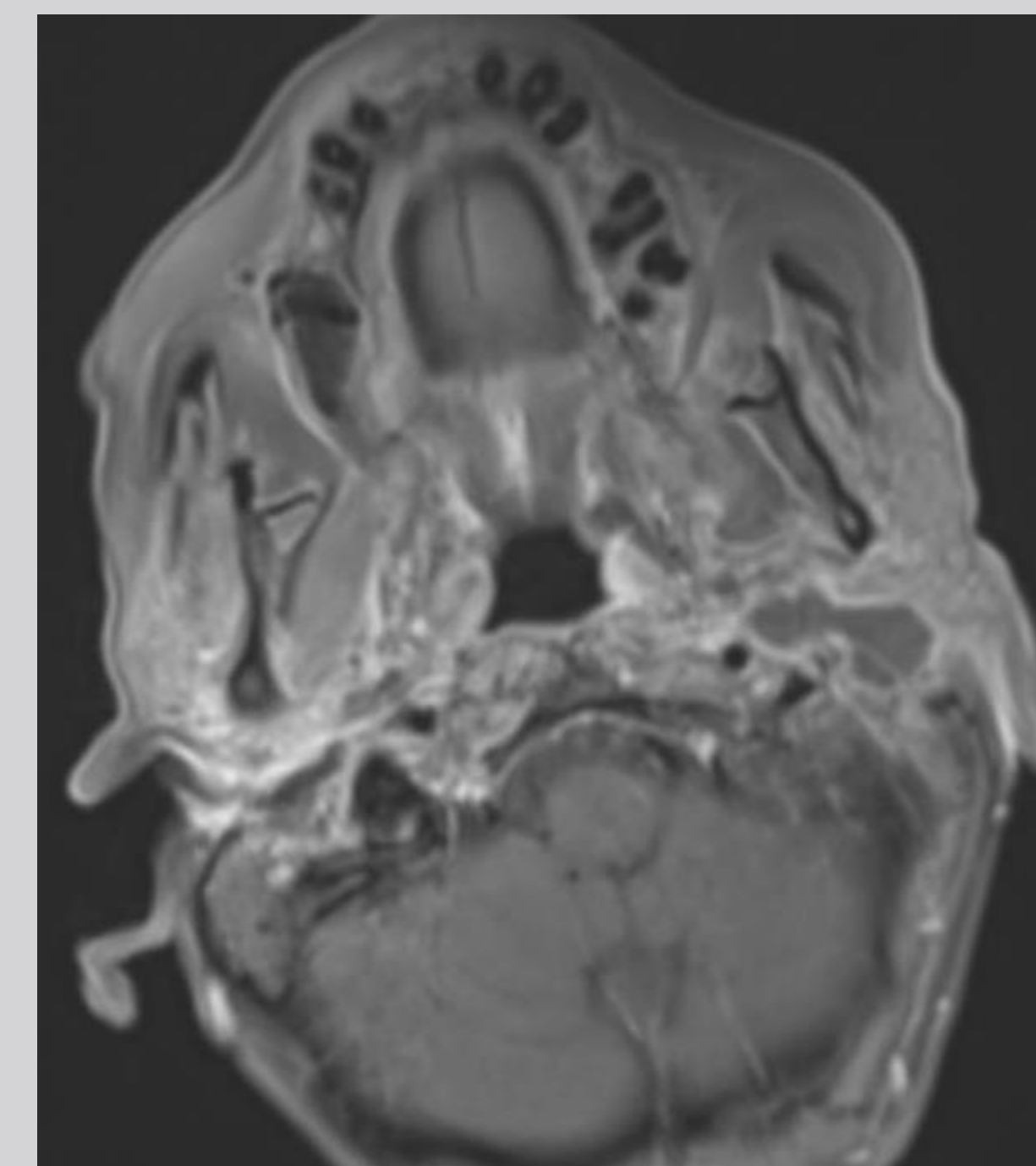


Figure 4. MRI neck of the patient in Case 2 demonstrating a 2.5cm left parapharyngeal space abscess.

CONCLUSION

In this report we have described two cases of malignant otitis externa caused by atypical organisms. In each case, the causative pathogen was eventually determined to be a gram-positive bacterium. The epidemiology of malignant otitis externa is evolving, and practitioners should consider gram-positive organisms in addition to *Pseudomonas* when initiating treatment for this condition. Medical treatment should be tailored to the individual patient based on culture data. In addition, one of the cases described had a positive outcome after being managed with tympanomastoidectomy. Although not the mainstay of treatment, practitioners should consider surgical therapy in patients with progressive chronic malignant otitis externa refractory to antimicrobial treatment.