

Case Report

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Refractory chronic otitis media and mixed hearing loss due to infection with levaquin-resistant *KERSTERSIA GYIORUM*: A case report and review of the literature

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Abstract

A 39-year-old Caucasian male, with a past medical history of purulent drainage of both ears as an infant and multiple tympanostomy tube placements in childhood, presented our ENT clinic complaining of right ear fluid drainage and hearing loss for the past year. Two months prior to presentation, he began to have increased fluid drainage from the right ear. Examination of the right external auditory canal showed thick, white fluid and a tympanic membrane perforation with a protruding small mass. A sample of the fluid was obtained. Culture of the right EAC fluid grew aerobic, gram-negative rods, identified as *Kerstersia Gyiorum*. The patient was initially treated with topical ofloxacin and topical prednisolone drops. He was seen again 4 weeks later and was noted to have persistent fluid drainage from the right ear. At this time, oral sulfamethoxazole-trimethoprim was added to his regimen.

Keywords: Chronic otitis media; hearing loss; treatment resistant otitis media; uncommon infectious microorganism; *Kerstersia Gyiorum*

Abbreviations: COM: Chronic Otitis Media; CSOM: Chronic Suppurative Otitis Media; EAC: External Auditory Canal; S: Susceptible; I: Intermediate; CSF: Cerebrospinal Fluid; WHO: World Health Organization.

Introduction

Chronic Otitis Media (COM) is characterized by fluid discharge through a tympanic membrane perforation for greater than 2-6 weeks [1]. A 2004 document authored by the WHO estimated the disease burden of chronic suppurative otitis media to involve between 65 to 330 million individuals worldwide [2]. The same study attributed around 28,000 annual fatalities to this disease [2]. COM is a common disease that can result in significant morbidity and mortality. Complications of COM can typically be classified as extracranial and intracranial. Extra-

cranial sequelae include conductive and sensorineural hearing loss, development of cholesteatomas, inflammatory tissue development, and more [3]. Although intracranial complications less commonly occur, sequela are typically more dangerous and range in severity from a simple brain abscess to cavernous sinus thrombosis [3]. The possibility of such serious complications due to COM further emphasizes the importance of astute recognition and proper treatment.

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Pseudomonas aeruginosa and *Staphylococcus aureus* are the most common causative organisms in cases of chronic suppurative otitis media [4]. Other microbes have been implicated in the development of chronic otitis media, though, and infections with uncommon organisms have been described. *Kerstersia Gyiorum* has recently been recognized as a rare but possible cause of chronic otitis media, with only a few cases having been reported worldwide [5]. While infection with *Kerstersia Gyiorum* is infrequent, it is important to recognize this microbe as a possible cause of COM that is resistant to conventional treatment. A study examining three separate cases of chronic suppurative otitis media caused by *Kerstersia Gyiorum* found that two of the three cases had isolates resistant to ciprofloxacin [6]. It is also important to note that *Kerstersia Gyiorum* can easily escape identification, as many commercial identification systems do not contain this microbe in their databases [6,7]. It is critical to recognize *Kerstersia Gyiorum* and its role in COM, as long-term complications, especially hearing loss, are more likely to affect these patients.

In this report, we describe a case of recurrent chronic otitis media with associated hearing loss caused by a rare organism (*Kerstersia Gyiorum*) that was resistant to primary treatment options (ciprofloxacin).

Case report

A 39-year-old Caucasian male presented to the otolaryngology clinic complaining of fluid drainage from his right ear for approximately one year in duration. The drainage was constant during this time and began to worsen two months prior to presentation. The patient reported subjective hearing loss on the right side but denied any similar symptoms of his left ear. At presentation, there was no dizziness, vertigo, tinnitus, or otalgia on either the right or left side. The patient also denied any constitutional symptoms, including fevers, chills, headaches, and weight changes.

According to the patient's mother, he began to experience significant purulent drainage from both ears at the age of 10 months. Importantly, he was unable to receive any form of medical treatment for this issue until the age of 2. The patient had a history of multiple tympanostomy tube placements during childhood, but the exact age(s) at which he received these tubes was unknown. The patient stated that he has had bothersome ears for the majority of his life.

The patient is from a small, rural town with a population of less than 6,000 people. Of note, he had a 10-pack-year smoking history and was still using cigarettes at the time of initial presentation. He denied any illicit drug use and reported alcohol consumption only at social events.

External examination of both ears and cranial nerve testing revealed no abnormalities. Hearing was grossly intact bilaterally. Using a tuning fork at 512 Hz, Weber and Rinne testing demonstrated appropriate air and bone conduction of both ears. Otomicroscopy was then performed. Inspection of the right ear revealed a significant amount of thick, white fluid present in the right external auditory canal. This fluid was removed and sent for culture. Following suction, the right external auditory canal was noted to be erythematous. Inspection of the right tym-

panic membrane revealed a perforation and the presence of a grape-like mass protruding through the perforated membrane. The mass was noted to be friable and soft upon palpation with the suction device. Otomicroscopy of the left external auditory canal showed no abnormalities and an intact tympanic membrane.

Prior to culture results, the patient was prescribed topical ofloxacin drops and prednisolone drops, with instructions to apply four drops twice daily of each medication. Approximately one week later, culture of the right external auditory canal aspirate identified *Kerstersia Gyiorum* as the causal microorganism.

The patient was seen one month later at our clinic. Repeat binocular microscopy of the right ear again showed significant purulence in the external auditory canal, although there was less fluid compared to the initial clinic visit. The mass protruding through the perforated tympanic membrane was still present, although it was noted to have decreased in size. Following this visit, the patient was prescribed oral sulfamethoxazole-trimethoprim 400-80 mg with instructions to take one tablet twice daily in addition to continuing topical use of ofloxacin and prednisolone drops. The patient was then scheduled for a CT scan of the right temporal bone and an audiogram at his follow up appointment six weeks later.

Following examination in the otolaryngology clinic, aspirate of the patient's right EAC fluid was sent for culture and identification. Gram stain of the aspirate showed few gram-positive cocci, few gram-positive rods, few gram-negative rods, and few gram-positive rods. No organisms were isolated on anaerobic culture. Aerobic culture isolated a moderate number of gram-negative rods, and further investigation identified *Kerstersia Gyiorum* as the causal microorganism. Fungal culture yielded no growth after four weeks.

Antibiotic susceptibility testing of the *Kerstersia Gyiorum* isolate yielded the following results:

Table 1: Antibiotic susceptibility of *Kerstersia Gyiorum* isolate

Antibiotic	MIC (µg/mL)	Interpretation
Piperacillin-Tazobactam	≤ 16/4	S
Cefepime	≤ 2	S
Ceftazidime	≤ 4	S
Meropenem	≤ 0.12	S
Aztreonam	8	S
Ciprofloxacin	2	I
Levofloxacin	1	S
Amikacin	≤ 8	S
Gentamicin	≤ 1	S
Tobramycin	≤ 1	S
TMP/SMX	≤ 0.5/9.5	S

(S: Susceptible; I: Intermediate).

The differential diagnosis for this patient who presented with a ten-month history of unilateral ear drainage and associated hearing loss included cerumen impaction versus otitis media versus cerebrospinal fluid leak. Initial history of present illness and physical exam were key factors in narrowing of this working differential. EAC did not contain significant amount of cerumen on inspection which effectively ruled out impaction as the cause of his symptoms. CSF leak was low on our differential due to the patient's lack of previous head trauma and EAC findings which were inconsistent with typical clear and watery appearance of CSF. The fluid seen in the right EAC was noted to be thick and white, an appearance most consistent with purulent fluid secondary to infection. This physical exam finding, in addition to a history which described a ten-month duration of symptoms, encouraged a diagnosis of chronic otitis media.

The small, grape-like mass protruding from the tympanic membrane could represent cholesteatoma or inflammatory granulation tissue. The patient's history of multiple tympanostomy tube placements, as well as chronic tympanic membrane perforation, favored the diagnosis of cholesteatoma. Following improvement in symptoms and return to our clinic, we will obtain CT imaging of his right temporal bone to identify the origin and extent of this small mass.

The patient is scheduled to return to the clinic in six weeks for follow up. At this time, he will be assessed for resolution of his infection. Plan of care for this encounter includes a CT scan of his right temporal bone and an audiogram of both ears.

Discussion

While otitis media is commonly seen in clinical practice, it is exceedingly rare to identify *Kerstersia Gyiorum* as the causative infectious organism. There have only been 6 reports of chronic otitis media due to this microorganism, with a total of 9 cases described worldwide [5-10]. Each of the cases identified *Kerstersia Gyiorum* as the cause of chronic otitis media, with seven of the nine cases describing Chronic Suppurative Otitis Media (CSOM) and one case describing cholesteatomas chronic otitis media. A report by Uysal and colleagues discussed a 25-year-old male with recurrent otorrhea since childhood who was found to have CSOM due to *Kerstersia Gyiorum* [9]. When cultured and tested for antibiotic susceptibility, the clinical isolate exhibited resistance to aztreonam, ciprofloxacin, and trimethoprim-sulfamethoxazole, with intermediate resistance to levofloxacin and ticarcillin-clavulanate [9]. In this patient, sensitivities revealed an intermediate susceptibility to ciprofloxacin and susceptibility to levofloxacin. Using the literature at hand, the decision was made to steer away from using fluoroquinolones in the treatment of *Kerstersia Gyiorum*.

This particular case adds to the few reported cases of this rare and potentially disabling cause of COM. Cases such as these demonstrate the importance of early recognition of uncommon causes of chronic otitis media. In cases of chronic otitis media that are refractory to initial pharmacotherapy, further investigation should be quickly executed. Furthermore, the long-term complications of chronic otitis media, especially hearing loss, may be reduced by prompt identification of the causal microorganism and appropriate medical and/or surgical therapy.

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