Case Report

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Shewanella algae in a chronic suppurative otitis media patient with cholesteatoma

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Summary **We present *Shewanella algea* infection in a chronic suppurative otitis media (CSOM) patient with cholesteatoma in terms of clinical course and treatment. This is the first time *S. algea* is found as solely pathogen in a CSOM patient without history of contact with seawater in Turkey. The patient admitted to the hospital several times with complaints of otorrhoea, was diagnosed as otitis media and treated. He was hospitalized to the otorhinolaryngology department for further evaluation of recurrent infections. The patient was diagnosed as cholesteatoma according to computed tomography scan findings and was operated for cholesteatoma. As a result of surgical and medical treatment he was discharged with full recovery. Physicians must be aware of rarely seen pathogens and their unexpected ways of transmission and underlying causes such as cholesteatoma when treating patients for CSOM.**

Keywords: Shewanella, chronic suppurative otitis media, cholesteatoma

1. Introduction

The genus *Shewanella* are widely distributed in the environment as motile, non-fermentative, facultative anaerobe, saprophytic Gram-negative bacilli. *Shewanella* spp. is a member of the marine microflora and is a rare pathogen for human being. The prevalence of Shewanella infection is high in geographic regions with temperate climates such as parts of United States, South Africa, Australia, Asia and Southern Europe (*1,2*). *Shewanella* infections are also seen in Turkey due to climate features. Direct contact to seawater or consumption of contaminated seafood are well-known risk factors for infections (*3*). We present *Shewanella algae* in a chronic suppurative otitis media (CSOM) patient with cholesteatoma without exposure to marine environment.

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2. Case Report

A 34 year-old male was admitted to otorhinolaryngology clinic on December 17, 2016 with otorrhea and hearing loss. The patient had complaints of intermittent ear discharge for a long time and the hearing loss occurred afterwards. He was treated twice in the outpatient clinic in 2015. In December 2016 he was admitted to the hospital with the same complaints. According to physical examination findings, the patient was diagnosed as otitis media. Ofloxacin 0.3% ear drop and a corticosteroid ear drop was prescribed. The patient did not recover despite medical treatment. He was hospitalized in the otorhinolaryngology department for further evaluation on January 4, 2017. Emprical treatment with ampicillin-sulbactam 1 g 3 × 1 (IV), ofloxacin 0.3% ear drop 1 g 3 × 1, corticosteroid 1 mg ear drop were started. Ciprofloxacin (IV) 200 mg 3 × 1 was added to the treatment of the patient who did not give response to treatment. The patient did not report any contact with seawater or marine environment. The biochemical test result of C-reactive protein was 3.3 mg/L (reference 0-3.5 mg/L). Complete blood count and erythrocyte sedimentation rate results were within normal limits.

Samples of middle-ear discharge were collected

from the patient, by an otorhinolaryngology specialist, under strict aseptic conditions using sterile swabs, after cleaning the external auditory canal with a different swab. The swab samples were immediately sent to the microbiology laboratory for bacterial studies. The swab sample was cultured on 5% sheep blood agar and eosin methylene blue agar for isolation of aerobic bacteria, and incubated aerobically at 37°C for 24-48 hours. The isolates grown were analyzed according to standard microbiological and biochemical methods. After 24 hours of incubation non-fermentative colorless colonies were observed on eosin methylene blue agar while hemolytic mucoid colonies were noted on sheep blood agar. According to biochemical tests these organisms were catalase, oxidase positive and H_2S production was positive. Urea was hydrolysed. The solely isolated organism from middle-ear discharge was identified as *Shewanella algae* by Vitek2 Compact (bioMerieux, France) automated identification system.

According to antimicrobial susceptibility results which were obtained from VITEK 2 Compact system; the strain was sensitive to ceftazidime, imipenem, meropenem, amikacin, gentamycin, and ciprofloxacin and resistant to trimethoprim/sulfamethoxazole, cefuroxime, ampicillin/sulbactam and amoxicillin/clavulanic acid.

In microscopic ear examination, purulent discharge, epithelium and cholesteatoma were seen. In the computed tomography mastoid cavity was also filled with soft tissue consistent with cholesteatoma. Due to the antibiotic susceptibility results, treatment was continued with ciprofloxacin (IV) 400 mg. As soon as ear discharge ended, patient with CSOM was operated for cholesteatoma on January 11, 2017. The patient was discharged with full recovery as a result of medical and surgical treatment. The written informed consent was obtained from the patient for this study.

3. Discussion

CSOM is formed by chronic inflammation of the middle ear and mastoid mucosa in which the tympanic membrane intactness (perforation or tympanostomy tube) was disturbed and otorrhea is usually present. However there is no consensus about the duration of the symptoms. CSOM in some studies is described as otorrhea through a perforated tympanic membrane continuing for at least 2 weeks, whereas other studies accept this period lasting for 2-6 weeks. It is thought that CSOM develop after an unsuccessfully treated acute otitis media infection (*4*). In our case, the patient also suffered from CSOM.

Moreover recurrent CSOM is due to one or a combination of several factors. These include therapy with oral antibiotics alone, treatment with non-antibacterial drops, uncompliance with the treatment process, infection with resistant bacteria such as *P. aeruginosa* or MRSA, and/or the presence of a cholesteatoma. Disease can not be managed and tends to be recurrent in patients with a disrupted ear anatomy or who are prone to infections (*5*). He was admitted to the hospital two times with ear discharge complaints in 2015. According to physical examination findings, the patient was diagnosed as otitis media and treated empirically. In December 2016, he was applied to the hospital with the same complaints and did not respond to treatment. He was hospitalized for further analysis to investigate the underlying cause of recurrent infections.

Cholesteatoma is a well-demarcated non-cancerous cystic lesion derived from an abnormal growth of keratinizing squamous epithelium in the temporal bone. Cholesteatoma results from the enzymatic activity of the cholesteatoma matrix. This abnormal growth is locally invasive and capable of causing the destruction of structures in the middle ear cleft (*6-8*). The growth of cholesteatomas often progress undetected for years until they have become dangerously large and threaten to invade intratemporal structures and cause intra- and extracranial complications (*9,10*). Recurrent or persistent otorrhea over a period of 2 weeks should be included in differential diagnosis as a possible warning sign of cholesteatoma, particularly when these symptoms persist despite treatment or in cases involving a suspicious hearing loss in an ear that has previously been operated on (*11*). Our patient had recurring otorrhea several times persisting for more than 2 weeks.

It has been reported that *Shewanella* spp. are responsible for a wide range of clinical conditions such as skin and soft tissue infections, bacteremia, gastroenteritis, cerebellar abscesses, ear and eye infections, infective endocarditis and pericarditis. Direct contact to seawater or consumption of contaminated seafood are well-known risk factors for infections (*1,3,12,13*). Pneumonia, cerebellar abscess, meningitis and wound infections were the reported cases from our country (*14-16*). Yılmaz *et al*. reported a case of a cerebellar abscess and meningitis caused by *Shewanella putrefaciens* and *Klebsiella pneumoniae* in a fisherman, secondary to chronic otitis media (*15*). In a Danish study, the most common ear infection was due to *S. algea* and 47 out of 55 patients suffered from clinical symptoms shortly after seawater contact (*3*). Numerous studies in the literature have confirmed the relationship between marine contact and disease, but this relationship has not been shown in some studies (*12,17*). To *et al*. indicated that the seawater contact cannot be documented by some studies because of some limitations such as being retrospective which probably lack information on seawater contact (*12*). In our case we were able to obtain information that there was no seawater contact from the patient. Furthermore, the *Shewanella* infection rates increase during summer due to contact with marine environment (*12*). However the patient in our

study was admitted to otorhinolaryngology clinic first time in winter.

Shewanella isolates are usually found to be susceptible to third and fourth-generation cephalosporins, fluoroquinolones, aminoglycosides, erythromycin and chloramphenicol in many studies whereas they are less susceptible to trimethoprim/sulfamethoxazole. Some studies reported variable susceptibility results to ampicillin and cephalosporins (*2,3,18*). In accordance with these studies our *S. algea* isolate was susceptible to ceftazidime, imipenem, meropenem, amikacin, gentamycin, and ciprofloxacin whereas resistant to trimethoprim/sulfamethoxazole, cefuroxime, ampicillin/sulbactam and amoxicillin/clavulanic acid. In our case, it is the first time that *S. algae* was isolated as a sole pathogen in a patient with CSOM without a prior history of contact with seawater. In conclusion, physicians must be aware of these rare pathogens and underlying causes such as cholesteatoma when prescribing antibiotics for rarely seen pathogens in CSOM.

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