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Case report

Meningitis caused by *Streptococcus pyogenes* triggered by malignant otitis externa: A case report

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ABSTRACT

A previously healthy 58-year-old male initially presented to a healthcare facility with otorrhea in his left ear, accompanied by a scab in both the auricle and ear canal, with a lesion protruding from the left cavity and oozing discharge (day 0). The following day, the patient was transferred to our facility with fever and altered consciousness. Imaging studies, including computed tomography and magnetic resonance imaging, revealed a soft tissue mass in the left maxillary, ethmoid and sphenoid sinuses, and diffuse pus within the subdural space over the bilateral frontoparietal regions. Cerebrospinal fluid (CSF) analysis showed elevated opening pressure, increased white blood cell count, predominance of polymorphonuclear leukocytes, elevated protein levels, and reduced glucose levels compared to the serum. *Streptococcus pyogenes* was identified in ear discharge, CSF, and blood cultures using matrix-assisted laser desorption ionization time-of-flight mass spectrometry. The presence of a papilloma in the left nasal cavity suggested that malignant otitis externa (MOE) served as the entry point for infection, leading to meningitis. The patient was treated with a combination of ceftriaxone and vancomycin, followed by ceftriaxone and cefotaxime, surgical resection of the tumor, and local debridement for infection control. To date, this is the first reported case of *S. pyogenes*-induced meningitis secondary to MOE. Although rare, the detection of *S. pyogenes* in otitis externa emphasizes the necessity of comprehensive evaluation of ear pathology to enable effectively source control as well as manage related infections and complications.

Introduction

Streptococcus pyogenes is a catalase-negative, gram-positive, β-hemolytic bacterium that causes various infections, with clinical manifestations ranging from mild to severe—potentially life-threatening [1]. The most frequent presentation of *S. pyogenes* infection is acute pharyngitis; however, it has also been implicated in skin and soft tissue infections, and more rarely, in bacteremia, osteomyelitis, pneumonia, otitis media, and sinusitis [2]. Although *S. pyogenes* is an infrequent cause of meningitis, it is associated with a high fatality rate when it does occur [3]. Early initiation of antimicrobial therapy is vital for optimizing patient outcomes, as *S. pyogenes* remains susceptible to various antimicrobial agents [4].

Malignant otitis externa (MOE), also known as necrotizing external otitis, is a progressive, invasive infection of the external auditory canal that extends to the skull base, leading to osteomyelitis. It predominantly affects elderly individuals with diabetes mellitus [5]. Although *S. pyogenes*-induced otitis externa has been reported [6,7], no cases of *S. pyogenes*-induced MOE have been documented as far as we searched. Otitis externa is a potential route of bacterial entry that may lead to meningitis. Moreover, a comprehensive literature review using PubMed revealed no cases of *S. pyogenes*-associated meningitis developed post-MOE. In this report, an unusual case of *S. pyogenes*-induced MOE triggering meningitis is presented.

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Case report

A previously healthy 58-year-old man initially presented to a healthcare facility with left-sided otorrhea, accompanied by a scab within both the auricle and ear canal. The lesion protruded from the ear cavity with exudative discharge (day 0) (Fig. 1). The absence of an otolaryngologist made providing appropriate care difficult. Therefore, the patient was scheduled to be referred to our hospital's otolaryngology department. No antibiotic treatment was administered at the previous hospital. The following day, the patient returned with a fever and an altered mental status, leading to his transfer to our hospital. On physical examination, he demonstrated impaired consciousness (Glasgow Coma Scale [GCS] E4V1M1), a body temperature of 39.0 °C, blood pressure of 150/88 mmHg, pulse rate of 89beats/min, respiratory rate of 12breaths/min, and oxygen saturation of 98 % in ambient air. Clinical findings included nuchal rigidity, thickening of the left ear canal wall, and white otorrhea. Laboratory results revealed an elevated white blood cell count (28,000/μL), increased alkaline phosphatase (240 U/L), C-reactive protein (CRP; 35.94 mg/dL), blood glucose (147 mg/dL), and HbA1c (6.7 %) (Table 1). Cranial computed tomography (CT) revealed a soft tissue mass in the maxillary, ethmoid and sphenoid sinuses, with no evidence of an intracranial mass (Fig. 2**-a)**. Magnetic resonance imaging (MRI) demonstrated diffuse pus with high signal intensity in the subdural space over bilateral frontoparietal regions (Fig. 2**-b)**. Lumbar puncture revealed cerebrospinal fluid (CSF) with a clear appearance, elevated opening pressure (300 mmH_2O), a white blood cell count of 367/μL, predominantly polymorphonuclear neutrophils (89.6 %), elevated protein levels (382 mg/dL), and low glucose levels (3 mg/dL) (Table 1). Owing to altered consciousness, the patient required intubation. Empirical treatment with intravenous vancomycin (2.25 g every 24 h), meropenem (2 g every 8 h), liposomal amphotericin B (5 mg/kg every 24 h), and dexamethasone (13.2 mg every 24 h) was initiated to treat community-acquired meningitis and mucormycosis (Fig. 1). Endoscopic sinus surgery was performed on the same day, and the nasal cavity tumor was excised and later identified as a papilloma by biopsy. After finding gram-positive cocci (GPC) was detected in the Gram staining of ear secretions and CSF (after centrifugation), meropenem and amphotericin B were discontinued, and ceftriaxone (2 g every 12 h) was added to the antibiotic regimen. On day 2, blood cultures from two sets confirmed GPC (chain) growth. Cultures from ear discharge, CSF, and blood were positive for *S. pyogenes*, as confirmed by matrix-assisted laser desorption/ionization time-of-flight mass spectrometry on day 5. Subsequently, vancomycin and dexamethasone were discontinued, and ceftriaxone was continued. On day 7, ceftriaxone was replaced with cefotaxime (2 g every 8 h), which was selected because of its blood-brain barrier penetration and the persistent liver dysfunction since admission, which led to the preference to avoid hepatic-metabolized antibiotics. The mental status gradually improved (GCS E4VtM6), and he was extubated on day 8. Despite this progress, a follow-up MRI (multi-planar reconstruction) on day 11 revealed that the subdural abscess had become more apparent (Fig. 2**-c)**. The patient continued to recover, with normalization of his white blood cell count and CRP levels (Fig. 1). Swelling in the left external auditory canal decreased, and ear secretions became nonpurulent post-irrigation. Ear irrigation was halted on day 16. Owing to the small size of the subdural abscess, no surgical drainage was performed, and intravenous antibiotics were continued. Follow-up CT on day 26 showed a reduction in the abscess size. Cefotaxime treatment was continued until day 44. After the completion of antibiotic therapy, rehabilitation was provided for transient higher brain dysfunction and dysphagia. By discharge, the patient had recovered to their pre-exacerbation condition. The patient was discharged on day 62, without sequelae or recurrence.

Discussion

This report describes a case of *S. pyogenes* meningitis secondary to MOE. To the best of our knowledge, this is the first documented case of *S. pyogenes*-associated MOE that resulted in meningitis. Several notable points warrant further investigation.

MOE is a progressive, invasive infection of the external auditory canal that can cause osteomyelitis of the skull base [5]. Unlike typical otitis externa, MOE is complicated by bone tissue involvement. *Pseudomonas aeruginosa* is the primary pathogen in MOE, isolated in 50–90 % of cases, and has been more extensively studied compared to other, less frequent microorganisms. MOE caused by *P. aeruginosa* predominantly affects older adults, diabetic, or immunocompromised individuals, with symptoms including persistent otalgia and chronic ear otorrhea [8]. The complications associated with *P. aeruginosa*-induced MOE include osteomyelitis, cranial nerve palsies—especially involving the facial nerve—meningitis, and brain abscesses. Other pathogens implicated in MOE include *Proteus mirabilis*, *Aspergillus fumigatus*, *Klebsiella* spp., and *Staphylococcus* spp. [9]. Although cases of *S. pyogenes*

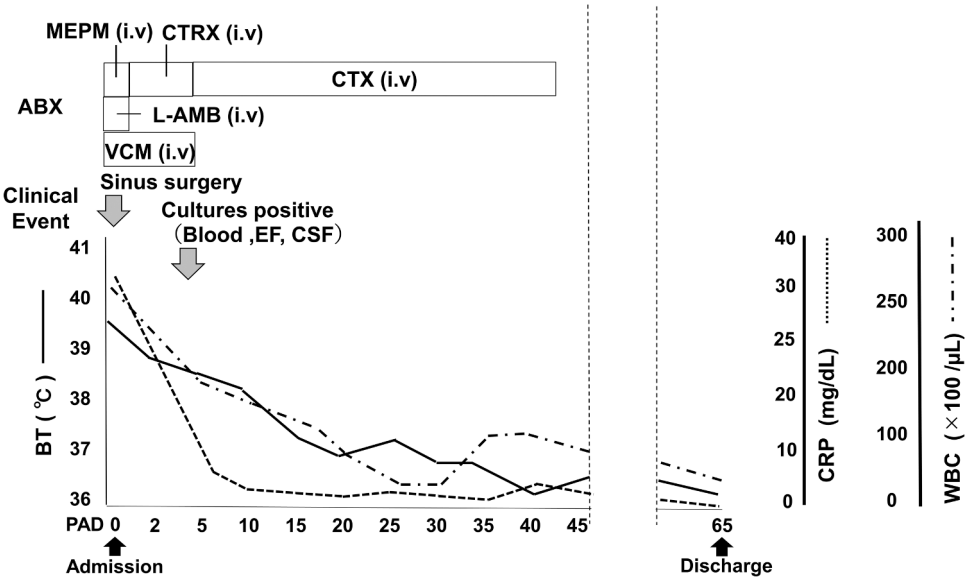

Fig. 1. Clinical course of the patient. ABX, antibiotics; MEPM, meropenem; CTRX, ceftriaxone; CTX, cefotaxime; L-AMB; Liposomal amphotericin B; VCM, vancomycin; IV, intravenous; EF, ear fluid; CSF, cerebrospinal fluid; BT, body temperature; CRP, C-reactive protein; WBC, white blood cells; PAD, post-admission day.

Table 1
Initial laboratory findings (blood and cerebrospinal fluid tests).

Blood test	Value	Normal range	Cerebrospinal fluid test	Value	Normal range
White blood cells (×1000/μL)	280	3.3–8.6	Opening pressure (cmH2O)	300	-
Neutrophils (%)	81	40–71.7	White blood cells (cells/mm3)	367	0–5
Lymphocytes (%)	13	28–44	Polynuclear sphere (%)	89.6	-
Monocytes (%)	6	4–8	Mononuclear sphere (%)	10.4	-
Red blood cells (×10000/μL)	453	435–555	Protein (mg/dL)	382	15–45
Hemoglobin (g/dL)	13.5	13.7–16.8	Lactate dehydrogenase	95	0–25
Hematocrit (%)	39.6	40.7–50.1	Glucose (mg/dL)	≤ 3	50–80
Platelets (×10,000/μL))	302	158–348	Albumin	1860	90–300
Sodium (mmol/L)	129	138–145	Lactic acid	96	0–25
Potassium (mmol/L)	4.2	3.6 - 4 .8			
Chloride (mmol/L)	91	101–108			
Albumin (g/dL)	3.4	4.1–5.1			
Total bilirubin (mg/dL)	1	0.4–1.5			
Blood urea nitrogen (mg/dL)	26	8–20			
Creatinine clearance (mg/dL)	1.13	0.65–1.07			
Urea nitrogen (mg/dL)	6.2	3–8			
Asparate transaminase (unit/L)	337	13–30			
Alanine transaminase (unit/L)	142	10–42			
Alkaline phosphatase (unit/L)	240	106–322			
Lactate Dehydrogenase (unit/L)	607	124–222			
C-reactive protein (mg/dL)	35.94	0–0.14			
Hemoglobin A1c (%)	6.7	4.6–6.2			
Activated partial thromboplastin time (s)	44.3	24–39			
Prothrombin time (s)	14.1	11–13			
Prothrombin time (%)	80	80–120			
Prothrombin time-international normalized ratio	1.15	0.9–1.1			

infection in otitis externa have been reported, no instances of *S. pyogenes*-induced MOE have been described. MOE treatment involves multiple surgical interventions, as demonstrated in this case, along with long-term monitoring of treatment response. Consequently, the involvement of otolaryngologists is crucial. Inadequate management of MOE can cause the infection to spread to the skull base, engendering severe sequelae, such as meningitis, thrombosis of the lateral sinus or internal jugular vein, Besold's abscess, and cranial nerve dysfunction [10].

S. pyogenes-induced meningitis is relatively rare, accounting for approximately 1 % of all invasive *S. pyogenes* infections [11] and comprises 0.2–1.2 % of all bacterial meningitis cases in both adults and children [12]. *S. pyogenes*-meningitis is frequently associated with concomitant otitis media (47 %) and mastoiditis (30 %) [13]. Herein, the papilloma in the nasal cavity may have disrupted the anatomical barriers, allowing the MOE to act as a portal of entry and facilitating the spread of *S. pyogenes* to the meninges. Therefore, in MOE diagnosis, a thorough assessment of possible meningitis extensions is essential. Similarly, when meningitis is suspected and ear-related symptoms are present, meticulous examination of the external ear and consideration of debridement of the otitis externa should be prioritized. As exemplified in this case, the detection of *S. pyogenes* as the causative pathogen in meningitis necessitates a comprehensive investigation of potential entry points, such as MOE.

In conclusion, this case report represents a rare instance of *S. pyogenes*-induced and MOE triggered meningitis. Papilloma in the left nasal cavity is instrumental in the disruption of normal anatomical defenses, thereby facilitating the spread of the infection. The patient was successfully treated with a regimen comprising ceftriaxone and vancomycin regimen, followed by ceftriaxone monotherapy and cefotaxime treatment. Surgical resection of the tumor and local debridement were performed to control localized infection. Although *S. pyogenes* is an uncommon cause of MOE, its identification as a pathogen in meningitis highlights the critical importance of a comprehensive evaluation of the external ear to identify potential sources of infection for effective source control.

Ethical approval

Written informed consent was obtained from the patient for the publication of this case report.

Consent

Written informed consent was obtained from the patient for the publication of this case report.

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Authorship statement

All authors satisfy the ICMJE authorship criteria. AS was involved in the treatment and contributed to writing the manuscript. DO was involved in the treatment and supervised the report. KM, KY, TK, YN, HO (Hideaki Ohno), and HO (Hideaki Oka) were involved in the treatment and provided feedback regarding clinical microbiology and treatment. KT provided feedback on clinical microbiology and treatment. ST was involved in the operation. MS was the doctor in charge of inpatient care. All authors approved the final version of this manuscript.

CRediT authorship contribution statement

Mimura Kazuyuki: Writing – original draft, Supervision. **Yamamoto Kei:** Writing – original draft, Supervision. **Kawamura Takayuki:** Writing – original draft, Supervision. **Tsukada Kunihisa:** Writing – original draft, Supervision. **Oka Hideaki:** Writing – original draft, Supervision. **Shirai Ayako:** Writing – original draft, Supervision. **Ono Daisuke:** Writing – original draft, Supervision. **Nozaki Yujin:** Writing – original draft, Supervision. **Tanaka Sunao:** Writing – original draft, Supervision. **Yamamoto Masaomi:** Writing – original draft, Supervision. **Ohno Hideaki:** Writing – original draft, Supervision.

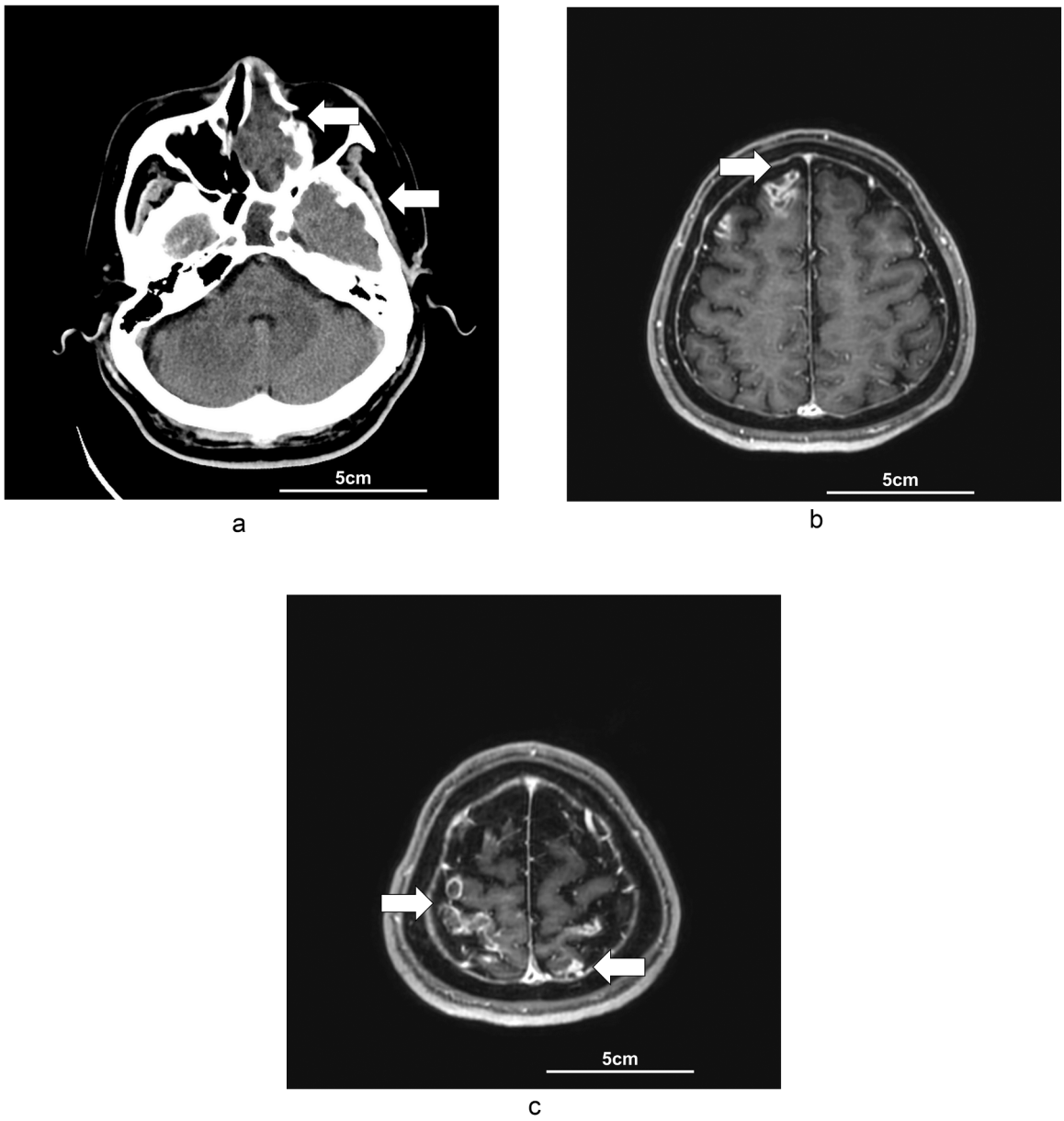

Fig. 2. Head computed tomography (CT) scanning and magnetic resonance imaging (MRI) findings. A CT scan on admission revealed a soft tissue shadow in the left maxillary, ethmoid and sphenoid sinuses, and did not show an intracranial mass (arrow; a). MRI revealed diffuse pus with high signal intensity in the subdural space over bilateral frontoparietal regions (arrow; b). MRI images (multiplanar reconstruction) revealed a subdural abscess on day 11 (arrow; c).

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper

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Conflicts of interest

None.

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