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RAOULTELLA ORNITHINOLYTICA IN A HEALTHY, YOUNG PERSON: RAPIDLY PROGRESSIVE SINUSITIS WITH ORBITAL AND INTRACRANIAL INVOLVEMENT

Jihyeon Baek[1], Marn Joon Park[2], Ha Young Lee[3], Sungmo Kang[4]

[1] Division of Infectious Diseases, Department of Internal Medicine, Inha University College of Medicine, Incheon, South Korea
[2] Department of Otorhinolaryngology-Head and Neck Surgery, Inha University College of Medicine, Incheon, South Korea
[3] Department of Radiology, Inha University College of Medicine, Incheon, South Korea
[4] Department of Ophthalmology, Inha University College of Medicine, Incheon, South Korea

Corresponding author: Jihyeon Baek **e-mail:** jhbaek@inha.ac.kr

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ABSTRACT

Raoultella ornithinolytica is an encapsulated, Gram-negative, nonmotile, rod belonging to the *Enterobacteriaceae* family. Infections involving the gastrointestinal tract and the hepatopancreatobiliary system are most frequently reported, especially in immunocompromised patients. The authors present an unusual case of acute complicated sinusitis with orbital and intracranial involvement caused by *R. ornithinolytica*. The infection was rapidly progressive, even though the patient was a healthy, young person without any co-morbidities. The patient's condition improved after antibiotic treatment and multiple ophthalmic and sinus surgeries.

KEYWORDS

Raoultella ornithinolytica, maxillary sinusitis, orbital cellulitis, abscess, meningitis

LEARNING POINTS

- *Raoultella ornithinolytica* can cause rapidly progressive infections, even in immunocompetent young individuals.
- *Raoultella ornithinolytica* identified in sinusitis can be a true pathogen rather than an innocent bystander.

CASE DESCRIPTION

The patient was a 22-year-old male. He did not have any co-morbidities or past medical history except for a febrile seizure at the age of 2 years. He visited the emergency room for pain and swelling of the right eye that had started 12 hours earlier. On the day of the visit, he had suffered from a headache that lasted for hours and he had vomited five times. He had also visited an ophthalmology clinic outside the hospital several hours before he presented to the emergency room and had been prescribed antibiotics. However, because the swelling progressed rapidly following this first clinic visit, he decided to present to the emergency room. He exhibited right ptosis, the right eyelid was swollen and erythematous, and supraduction was limited. He was also experiencing vertical diplopia with supraversion. Blood analysis revealed a white blood cell count of 12.00×10^9/l

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(normal range 4.00–10.00×10^9/l), with 80.1% neutrophils, an elevated C-reactive protein (CRP) level of 6.67 mg/dl (normal range 0.0–0.5 mg/dl) and an elevated erythrocyte sedimentation rate of 26 mm/hour (normal range 1–15 mm/hour). Orbit computed tomography showed fluid collection with mucoperiosteal thickening in the right frontal, ethmoid and maxillary sinuses, diffuse swelling of the right orbit and a subperiosteal abscess in the roof of the right orbit *(Fig. 1)*.

After the right middle meatal culture and blood culture on day 1, the patient was started on parenteral ampicillin/sulbactam and gentamicin. An orbitotomy for abscess drainage, an endoscopic middle meatal antrostomy and frontal sinusotomy were performed on day 3. Additional cultures were performed with pus from the middle meatus upon uncinectomy. The mucosal swelling and pyocele of the inferior portion of the right maxillary sinus suggested an odontogenic origin of infection. Considering the pachymeningitis and subdural empyema on brain magnetic resonance imaging (MRI), the antibiotics therapy was changed to ceftriaxone with metronidazole on day 5. As the several cultures from the right middle meatus revealed few *R. ornithinolytica* with or without few *Streptococcus intermedius*, ceftriaxone with metronidazole was continued as definite therapy *(Table 1)*. The blood culture revealed no bacterial growth. The patient was treated with ceftriaxone for a total of 4 weeks and metronidazole for 9 days, until his discharge. The second orbitotomy for abscess drainage and a revision endoscopic sinus surgery were performed on day 11 for the subperiosteal abscess, and the sinusitis persisted. The maxillary second molar and wisdom teeth were surgically extracted for severe dental carries and an apical abscess on day 15. The patient was discharged on day 34 after the resolution of his epidural empyema and pachymeningeal enhancement had been confirmed, and

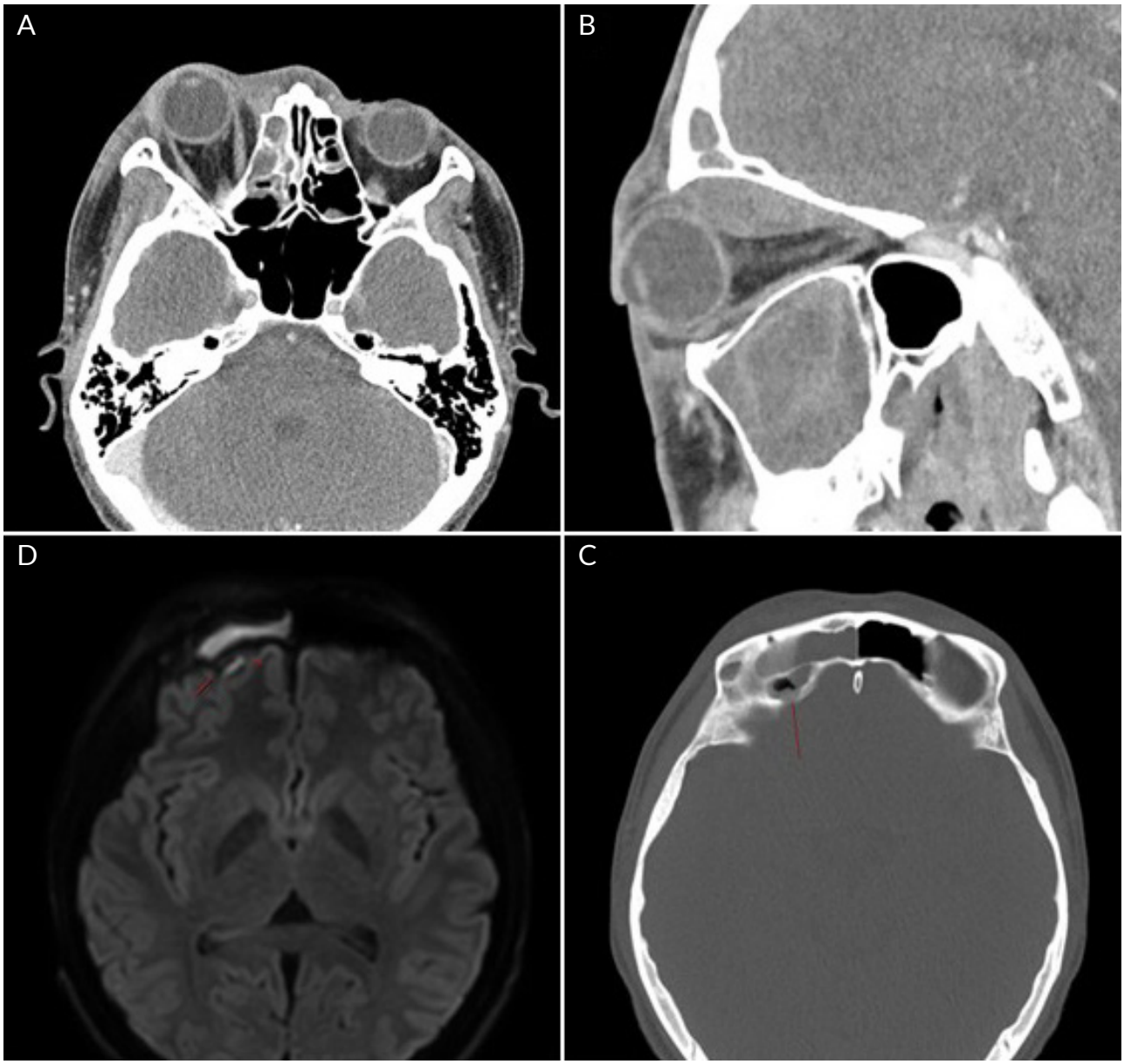

Figure 1. Representative images from axial and sagittal and bone setting images of post contrast CT (A, B, C) and a B-1000 image of diffusion weighted imaging brain magnetic resonance imaging (D), Clockwise from top left: A. Proptosis of right eye, B. Abscess pocket in right superior orbit, Acute sinusitis in right frontal and maxillary sinuses, C. Erosion in inner wall of right frontal sinus, D. Minimal epidural abscess in right frontal convexity.

Raoultella ornithinolytica	
Antibiotic	*Classification*
Amikacin	Susceptible (MIC ≤ 2)
Ampicillin	Resistant (MIC = 16)
Aztreonam	Susceptible (MIC ≤ 1)
Cefazolin	Susceptible (MIC ≤ 4)
Cefotaxime	Susceptible (MIC ≤ 1)
Cefoxitin	Susceptible (MIC ≤ 4)
Ceftazidime	Susceptible (MIC ≤ 1)
Cefepime	Susceptible (MIC ≤ 1)
Ciprofloxacin	Intermediate (MIC = 0.5)
Gentamicin	Susceptible (MIC ≤ 1)
Imipenem	Susceptible (MIC ≤ 0.25)
Piperacillin-tazobactam	Susceptible (MIC ≤ 4)
Trimethoprim-sulfamethoxazole	Susceptible (MIC ≤ 20)
Amoxicillin-clavulanic acid	Susceptible (MIC ≤ 2)
Ertanenem	Susceptible (MIC ≤ 0.5)
Tigecycline	Susceptible (MIC ≤ 0.5)
Streptococcus intermedius	
Antibiotic	*Classification*
Cefotaxime	Susceptible
Cefepime	Susceptible
Clindamycin	Susceptible
Erythromycin	Susceptible
Vancomycin	Susceptible

Table 1. Results of antibiotic susceptibility testing.

brain MRI had shown a marked improvement of the orbital abscess. Persistent right maxillary sinusitis due to drainage problems for mucosal oedema was detected after discharge and treated with additionally prescribed oral amoxicillin-clavulanate. The patient improved with the prolonged oral antibiotic therapy, which was continued for 4 weeks, and did not require additional surgery.

DISCUSSION

Raoultella ornithinolytica is an encapsulated, Gram-negative, nonmotile rod that belongs to the family Enterobacteriaceae. It is distinguished phylogenetically from the genus *Klebsiella* and reclassified, as the genus *Raoultella*, with two other species, *Raoultella planticola* and *Raoultella terrigena*. *Raoultella ornithinolytica* can cause histamine poisoning with cutaneous flushing, known as scombroid syndrome, which often occurs as a result of fish poisoning. Human infections caused by *R. ornithinolytica* are rare, and their clinical characteristics are not fully known. Recent reports have pointed to a role of *R. ornithinolytica* in human infections, mainly in immunocompromised patients, and some multi-drug-resistant strains have been identified[1].

Many of the previously reported human cases of *R. ornithinolytica* infections occurred in patients with impaired immunity due to malignancy, immunosuppressants or old age. Most infections in healthy, young individuals are healthcare-associated or related to trauma. The gastrointestinal tract and the hepatopancreatobiliary system are the most frequently reported infection sites, but urinary tract and osteoarticular infections have also been reported[1]. Very few cases of sinusitis with *R. ornithinolytica* have been reported[2,3]; in these cases, *R. ornithinolytica* caused severe pansinusitis or spread to adjacent facial structures. In our patient, the odontogenic sinusitis rapidly spread and caused orbit cellulitis, a subperiosteal abscess, pachymeningitis and intracranial epidural empyema. To the best of our knowledge, this is the first case of sinusitis complicated by *R. ornithinolytica* with orbital and intracranial involvement. The patient did not experience any symptoms until 12 hours prior to his presentation, and his symptoms rapidly deteriorated over the course of several hours. Laboratory investigations were performed to explain the rapid progress in this patient, but no deficits in humoral and cellular immunity were observed. The authors identified other reports of rapidly progressive community-acquired infections in a healthy person caused by *R. ornithinolytica*[4].

The fact that *R. ornithinolytica* infections can progress rapidly in immunocompetent, young people suggests that this rare human pathogen has unidentified virulence factors, similar to other Enterobacterales. Earlier studies have also identified virulence genes in *R. ornithinolytica*[5]. Our patient showed a slow response to antibiotics, despite the antimicrobial susceptibility tests indicating susceptibility, which resulted in repeated ophthalmic and sinus surgeries during hospitalisation. Moreover, prolonged antibiotic treatment was required after discharge until sufficient improvement was observed. The virulence of the rare pathogen may explain the poor response to the antibiotic treatment.

These findings emphasise that *R. ornithinolytica*, a rare human pathogen, can be virulent not only in the immunocompromised but also in immunocompetent individuals. In the case presented here, a *R. ornithinolytica* infection with very little resistance was treated successfully with antibiotics and repeated surgeries. Considering its potential virulence and resistance, *R. ornithinolytica* identified as a cause of infection always requires susceptibility tests and careful treatment.

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