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Case report

A rare culprit: Ralstonia pickettii in a deep neck space infection

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ABSTRACT

Ralstonia pickettii is a rare, emerging opportunistic pathogen that has been previously limited to nosocomial infections, often associated with contaminated sterile solutions. Here, we present the case of a neck abscess caused by *R. pickettii*, the first documented case of a deep neck space infection caused by this bacterium. The patient in this case had no risk factors for *R. pickettii* infection. By highlighting the atypical presentation and microbiology in this case, we aim to highlight the emergence of a wide spectrum of disease caused by *R. pickettii*.

Introduction

Ralstonia pickettii, previously known as Burkholderia pickettii or Pseudomonas pickettii, is a gram-negative rod that is a low-virulent emerging opportunistic pathogen [1]. Within the Ralstonia genus, R. pickettii is the most likely to cause human infection. Originally found in water and soil, R. pickettii has been well-associated with nosocomial bacteremia and septicemia caused by contaminated fluid and sterile drug solutions [2-5]. Since the start of the COVID-19 pandemic, there has been an increase in reported R. pickettii infections, with numerous cases of bacteremia and peritoneal dialysis-associated infection described [6,7]. While there are no formal mortality rate estimates for infection, five of 29 (26.3%) patients associated with a contaminated hydromorphone outbreak in Colombia died, and there have been several other cases of death due to R. pickettii infection, making appropriate and early treatment of infection essential [2,3,8]. Here, we present the case of a neck abscess caused by R. pickettii, the first documented case of a deep neck space infection caused by this bacterium.

Case

A 72-year-old female with a history of hypertension, dyslipidemia, stage 2 chronic kidney disease, and type II diabetes mellitus presented for acute onset neck pain and swelling for the past three days. She stated that the pain originated under her left mandible and radiated to her left neck and chest. The patient had no pertinent risk factors for deep neck space infection and denied any recent dental manipulation, foreign body aspiration, trauma to the oral cavity or pharynx, intravenous (IV) drug use, tobacco use, or recent infections.

Upon presentation, the patient was afebrile with vital signs within normal limits. Physical exam was notable for a 3 cm area of induration under the angle of her left mandible with severe tenderness to palpation and pain with neck range of motion. There was no purulent drainage, and an ears, nose, and throat exam revealed normal dentition. The remainder of her physical exam was unremarkable.

Initial labs revealed an elevated white blood cell count of 14.6 K/uL, hemoglobin A1c of 12.2 %, and lactate level of 4.2 mmol/L, raising concern for deep neck space infection. Differential diagnosis included infected sialocele versus lymphadenitis versus malignancy. Contrastenhanced computer tomography (CT) of the neck revealed a rimenhancing fluid collection measuring $2.5\times2.1\times2.0$ cm beneath the left mandibular angle, invaginating and possible arising from the left parotid tail (Fig. 1). Associated thickening of the adjacent platysma and subcutaneous fat stranding were seen on imaging. There were no pertinent vasculature findings on CT imaging of the neck. Given the patient's clinical history, these findings raised concern for neck abscess, though cystic neoplasm could not be definitively excluded.

Empiric treatment with 3 g of IV ampicillin-sulbactam every six hours and 500 milligrams of ciprofloxacin once daily was started. The patient then underwent ultrasound-guided aspiration that yielded thick hemorrhagic clot-like material without frank purulence.

Over the next two days, the patient continued to endorse exquisite tenderness to palpation over the left angle of the mandible. Her white blood cell count peaked at 17.4 K/uL, and a repeat CT scan of the neck on hospital day three was unchanged from that on admission. At this time, initial wound cultures from the aspiration attempt on admission resulted in 1+ and 2+ growth of *Ralstonia pickettii*. Mistakenly thought to be a contaminant, antimicrobial sensitivities were not completed per

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laboratory protocol.

In the setting of the patient's progressive pain and delayed response to antibiotics, the head and neck surgery team performed an incision and drainage procedure on hospital day four. The inability to express any purulence raised concern for an underlying malignancy. A Penrose drain was placed, and cytology and culture of the bloody aspirate were sent to pathology, along with tissue samples of the neck mass for fresh and permanent sections.

On hospital day five, the patient's white blood cell count decreased to 6.7 K/uL. Although the patient continued to endorse moderate pain, there was minimal drainage at the incision site, so the Penrose drain that was placed the previous day was removed. The patient was discharged home with a 14-day course of 875 mg-125 mg amoxicillin-clavulanate every 12 h and close outpatient follow-up.

Several days after the patient was discharged, the wound culture from the incision and drainage resulted with 1+ gram negative rods, consistent with the previous culture growing R. pickettii. No further speciation or antibiotic sensitivity was performed. Pathology results later revealed salivary gland tissue, mature fibroadipose tissue, and skeletal muscle with acute and chronic inflammation, abscess formation, and necrosis. There was no evidence of malignancy. Six weeks after discharge, the patient reported complete resolution of symptoms.

Discussion

The case presented here is the first documented case of a deep neck space infection caused by *Ralstonia pickettii*. Neck abscesses and deep neck space infections are serious but relatively common conditions that often arise from the local extension of infections from odontogenic and other upper airway structures [9]. Typically, these infections are polymicrobial, with microorganisms from the oropharyngeal flora frequently identified. Diabetes mellitus is a known risk factor for deep neck infections, and pathogenic bacteria found in patients with diabetes more commonly includes *Klebsiella pneumoniae*, *Staphylococcus spp.*, and *Streptococcus spp* [10–12].

The presence of *R. pickettii* as the sole pathogen in the case discussed here is highly unusual. While it remains unknown exactly how and where the patient was exposed to *R. pickettii*, interestingly, its origin was not nosocomial or via contaminated fluid solutions in a hospital setting, as is often the case [1]. Previously identified risk factors for *R. pickettii* infection include the presence of a central line, end stage renal disease

with dialysis-dependence, and cystic fibrosis [6,13–15]. In this case, the patient's only predisposing factor was sub-optimally controlled type II diabetes mellitus, also lacking other traditional risk factors for a neck abscess [16]. Thus, this case broadens the clinical spectrum of disease caused by *R. pickettii* and underscores the importance of recognizing the potential of this microorganism to be the causative agent in atypical infections.

The initial misidentification of *R. pickettii* as a contaminant posed a significant diagnostic challenge. The patient's history of acute, progressive neck pain and swelling, elevated white blood cell count, and CT imaging strongly suggested a bacterial neck abscess. Empiric treatment with combination ampicillin-sulbactam and ciprofloxacin was started, per the antimicrobial stewardship guidelines at our institution for deep neck space infections. Per our institution's laboratory protocol, minimum inhibitory concentrations (MIC) and antibiotic sensitivities were not performed, despite *R. pickettii* growth in both of the initial wound cultures collected. Looking ahead, we urge clinicians to be discerning in evaluating whether *R. pickettii* is a true sample contaminant. Notably, Ralstonia species may require more than 72 h of incubation to visualize cultured colonies and can often also be misidentified as *Burkholderia cepacia* due to closely-related phylogenetics [17].

In cases such as this one, where there is no source of contamination, *R. pickettii* should be considered the causative pathogen, especially as the range of its infection potential continues to evolve. MIC should also be completed as part of the standard work-up to gain a better understanding of the resistance patterns of this emerging pathogen. There is currently no standard recommendation for treatment of *R. pickettii* infection. However, previous studies suggest that trimethoprim-sulfamethoxazole, fluoroquinolones, piperacillin-tazobactam, and the combination of a cephalosporin and aminoglycoside may be effective antimicrobial agents [6,14,18].

While originally low on the differential, suspicion of underlying malignancy rose significantly during the patient's hospitalization. The discrepancy between the rim-enhancing fluid collection seen on imaging and the absence of purulence and clear tissue planes appreciated intraoperatively was unexpected. Moreover, despite early initiation of broad-spectrum antibiotics, the patient continued to endorse worsening pain throughout the first several days of hospitalization with ongoing swelling and induration. While neck abscess as the initial presentation of malignancy is rare, it has been previously documented [19,20]. Therefore, clinical suspicion for malignancy should remain high, as deep neck



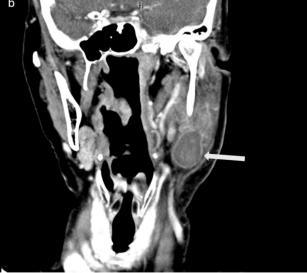


Fig. 1. Computer tomography scan of the neck in axial (a) and coronal (b) views demonstrating a rim-enhancing fluid collection. The lesion showed in these images is $2.5 \times 2.1 \times 2.0$ cm in size and consistent with neck abscess.

infection and malignancy are not mutually exclusive diagnoses.

In summary, we present the case of a neck abscess caused by *Ralstonia pickettii*, an emerging opportunistic pathogen that was previously limited to nosocomial settings. When identified on culture, *R. pickettii* should be considered a causative pathogen in infections even without traditional exposure risk factors or a clear source of contamination. Routine performance of MIC and antibiotic sensitivities should be conducted to ensure appropriate and early treatment. Use of trimethoprim-sulfamethoxazole, fluoroquinolones, piperacillin-tazobactam, or the combination of a cephalosporin and aminoglycoside may be considered as first-line agents, although no standard recommendation currently exists.

Author contribution

All authors were involved in patient care. All authors contributed to review of literature and manuscript writing. All authors have seen and approve of the final manuscript version that is being submitted. This manuscript is original work that has not been submitted or published elsewhere.

CRediT authorship contribution statement

Tara Shahrvini: Investigation, Writing – original draft, Writing – review & editing. **Matthew McCullough:** Investigation, Writing – original draft, Writing – review & editing.

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Ethical Approval

Institutional review board approval did not apply to this case.

Consent

All patient information has been deidentified to ensure patient privacy.

Conflict of Interest

None of the authors have conflicts of interest or declarations of interest to disclose.

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