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Case report

Rat bite fever caused by *Streptobacillus notomytis* mimicking pyogenic polyarthritis: A case report

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ABSTRACT

We report the world's sixth case of rat bite fever caused by *Streptobacillus notomytis* that mimicked pyogenic polyarthritis and required surgical debridement in combination with prolonged antibiotic therapy. This case report highlights the higher severity of rat bite fever caused by *S. notomytis* compared to *S. moniliformis*.

Introduction

Rat bite fever is a rare disease that is found mainly in Asian countries [1–5]. The classical manifestations include acute relapsing fever, rash, and migratory polyarthralgia [1,6]. This zoonotic disease can be transmitted through a rat bite or exposure to other animal vectors [7]. It is mainly caused by *Streptobacillus moniliformis* (gram-negative bacteria) or *Spirillum minus* (spirochete).

Streptobacillus notomytis was first isolated in 1979 from the heart of septicemic spinifex hopping mouse [7]. In 2018, the first human case of rat bite fever caused by *S. notomytis* was reported in Okinawa, Japan [1]. Until now, there were only five case reports of rat bite fever caused by *S. notomytis* [1–5]. *Streptobacillus* sp. is a fastidious gram-negative nonmotile bacillus that needs microaerophilic conditions to grow. The pathogen is unlikely to be identified by conventional methods [1,5–9]. Furthermore, sodium polyanethol sulfonate (SPS), an anticoagulant commonly used in the blood culture bottle was reported to have inhibitory effect with *Streptobacillus moniliformis* [10]. Therefore, rat bite fever may be underdiagnosed because it requires molecular diagnostic testing.

We report a case of rat bite fever caused by *S. notomytis* that mimicked pyogenic polyarthritis and required surgical debridement in combination with prolonged antibiotic therapy.

Case presentation

A 59-year-old man presented with persistent fever for 12 days before hospitalization. He had no significant medical history. He denied any contact with rats, other animal exposures, unpasteurized milk, or contaminated water. Twelve days earlier, he initially experienced high-grade fever without any localizing symptom. On day 2 of fever, he developed pain and swelling in both knees and ankles, mostly painful in the right ankle. The fever and joint pain partially subsided after acetaminophen and ibuprofen treatment.

On day 3 of fever, he had bilaterally painful and swollen wrists, knees, and ankles. He also noticed generalized nonpruritic erythematous maculopapular rashes around both knees and ankles (as shown in Fig. 1A). These rashes completely disappeared within 48 h.

On day 5 of fever, he visited Siriraj Hospital. Physical examination revealed a body temperature of 36.5 °C, bilateral symmetric polyarthritis of the wrists, knees, and ankles, without any skin lesions. Complete blood count (CBC) showed 8.760 WBC / uL with 78% neutrophils. The C-reactive protein was 171 mg/L. Dengue NS-1, Dengue IgG / IgM, and Chikungunya PCR were all negative. He was diagnosed with polyarthritis and had been treated with acetaminophen and tramadol. After treatment, high-grade fever and pain in all joints except the right knee partially improved.

On day 9 of fever, he had a follow-up visit at Siriraj Hospital. Physical examination revealed a body temperature of 36.8 °C, signs of mild

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arthritis of the wrists, left knee, and both ankles. The right knee had marked swelling and limited range of motion due to severe pain. CBC showed WBC 16,240/uL with 90.5% of neutrophils. The C-reactive protein was 181 mg/L. He was treated with naproxen.

On day 12 of fever, he still had severe pain in his right knee and persistent fever. He was later admitted to Siriraj Hospital. The presumptive diagnosis was pyogenic polyarthritis. The plain film of both knees revealed that the joint space was not narrowed and there were no significant abnormalities. The CBC showed WBC 13,310/uL with 84.7% of neutrophils and CRP 181 mg/L. Diagnostic arthrocentesis was performed that revealed yellow turbid synovial fluid. The synovial fluid WBC count was 13,700 with 71% neutrophils. Gram, AFB, modified AFB staining and crystal were negative. He was empirically treated with 2 g of ceftriaxone intravenously daily. Subsequently, therapeutic arthrocentesis was performed daily for 4 consecutive days. The synovial fluid WBC count varied between 13,700 and 113,600 cells / ml (neutrophils varied between 71% and 94%). After one week of ceftriaxone therapy, the signs of arthritis of both wrists, left knee, and both ankles recovered completely. However, fever and signs of arthritis of the right knee were still persistent. Both the synovial fluid culture and the automated blood culture (with SPS) later developed no organism. Ultrasonography of the right knee revealed bursal effusion along the lateral aspect of the suprapatellar bursa, measuring approximately 7.1 × 3.1 × 6.5 cm, with some internal septation, heterogeneous echoic content, and surrounding synovial thickening.

On day 23 of fever (day 10 of ceftriaxone therapy), arthroscopic debridement of the right knee was performed. The synovial biopsy of the right knee is shown in Fig. 1C-1D. The pathology report revealed acute synovitis, 20–50 stromal neutrophils per high-power field, presence of fibrinopurulent exudate, absence of crystal deposition, and negative for organisms, tumor, and granuloma. Three days after debridement, the fever completely subsided. Ceftriaxone was discontinued after the fever had completely gone for 5 days. The 16S rRNA gene PCR amplification finally reported the identification of *Streptobacillus notomytis*.

Discussion and conclusions

Streptococcus notomytis is a rare cause of rat bite fever. The classic manifestations of rate bite fever are acute relapsing fever, rash, and migratory polyarthralgia [1,6]. Approximately 50% of patients had migratory polyarthralgia [7]. However, arthritis with pyogenic synovial fluid is rare. In our patient, the clinical course and the profile of the synovial fluid mimicked pyogenic polyarthritis. The maximum WBC count of the synovial fluid from the right knee was 113,600/uL with 95% neutrophils, which was remarkable. Even without previous antimicrobial therapy, we were unable to identify the causative pathogen in both synovial fluid culture and blood culture. After ceftriaxone therapy, polyarthritis in all joints except the right knee joint improved significantly. The right knee arthritis did not respond to antimicrobial therapy and finally required surgical drainage. The confirmed diagnosis was carried out successfully using the molecular method.

Based on these findings, rat bite fever should be one of the differential diagnoses of culture-negative pyogenic polyarthritis among the population in an endemic area. Furthermore, additional surgical drainage may be needed among those who do not respond well to effective antimicrobial therapy. This case highlights the higher severity

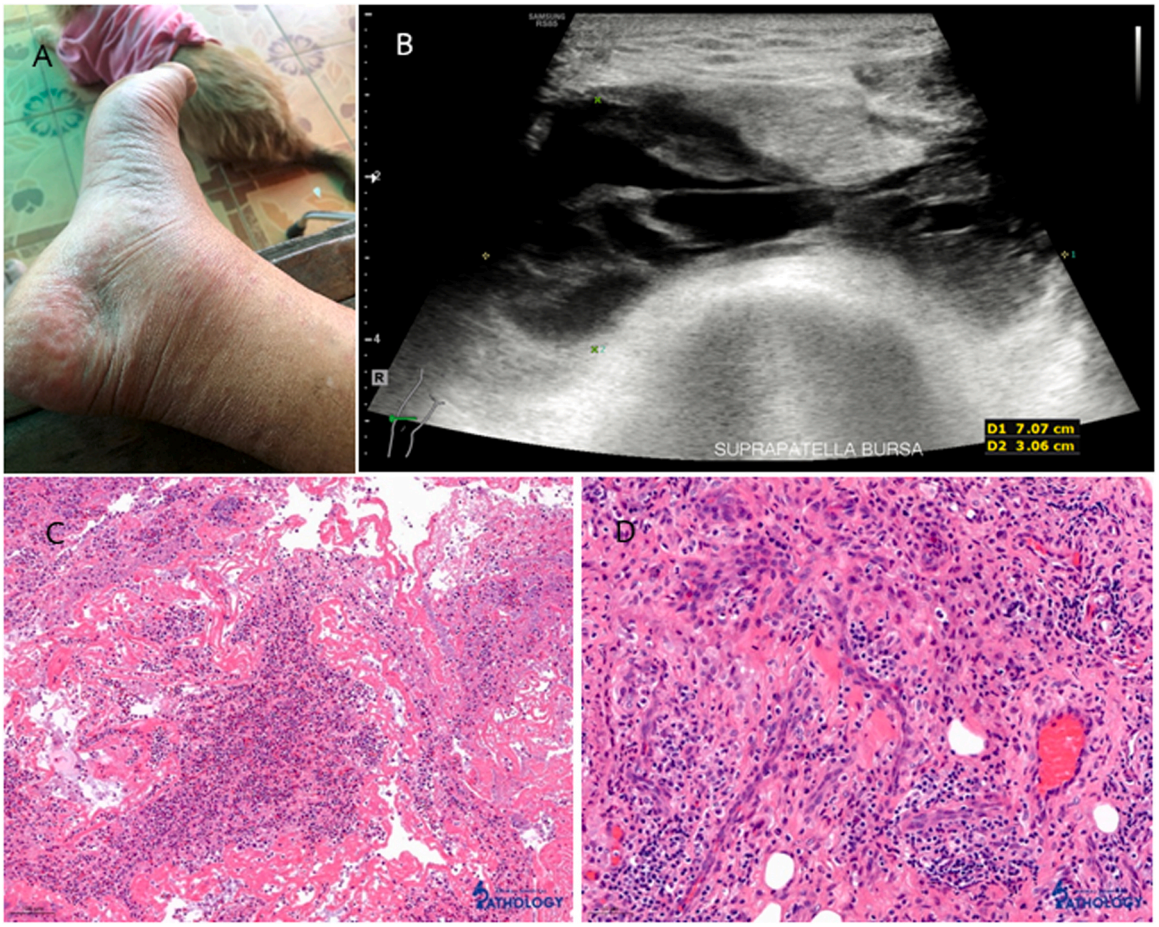

Fig. 1. (A) Non-pruritic maculopapular rashes in the right ankle; (B) Ultrasound of the right knee on day 19 of fever showing bursal effusion along the lateral aspect of the suprapatellar bursa, measured approximately 7.1 × 3.1 × 6.5 cm, with some internal septation, heterogeneous echoic content and surrounding synovial thickening; (C, D) H&E staining revealing acute synovitis with presence of fibrinopurulent exudate, absence of crystal deposition or granuloma.

of rat bite fever caused by *S. notomytis* compared to *S. moniliformis*.

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Consent

Informed consent was obtained from the patient for publication of this case report and accompanying images.

Ethical approval

The authors confirm that they have read and complied with the policy on ethical consent.

Author statement

We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship.

Author contributions

Patient treatment: NP, EA, SM, PR. Drafting the manuscript: NP, PR. All authors have read and approved the final version of the manuscript.

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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