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## Case Report

Unilateral inguinal lymphadenitis caused by *Yersinia pseudotuberculosis*. A case report<sup>☆</sup>Kentaro Iwata<sup>a,\*</sup>, Naomi Morishita<sup>b</sup>, Yoshiyuki Masuda<sup>c</sup>, Maki Kodama<sup>d</sup>, Sachiyo Otani<sup>d</sup>, Ayaka Naito<sup>d</sup><sup>a</sup> Division of Infectious Diseases Therapeutics, Kobe University Graduate School of Medicine, Kobe, Japan<sup>b</sup> Department of Nursing, Hyogo Prefectural Kakogawa Medical Center, Kakogawa, Japan<sup>c</sup> Department of Dermatology, Hyogo Prefectural Kakogawa Medical Center, Kakogawa, Japan<sup>d</sup> Department of Laboratory Medicine, Hyogo Prefectural Kakogawa Medical Center, Kakogawa, Japan

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## ABSTRACT

Acute inguinal lymphadenitis is usually caused by lower extremity infection and sexually transmitted diseases, such as chancroid, lymphogranuloma venereum, genital herpes, or syphilis. *Yersinia pseudotuberculosis* is a non-spore forming, pleomorphic, non-lactose fermenting Gram negative bacillus and a member of the family Enterobacteriaceae, which is associated with diarrheal diseases. It also causes mesenteric lymphadenitis at the terminal ileum, which can be clinically indistinguishable from acute appendicitis (pseudoappendicitis). However, lymphadenitis in other regions caused by the organism is rarely reported. Herein, we report a case of a man in his 20s, who presented with unilateral inguinal lymphadenitis caused by *Y. pseudotuberculosis*, with discussion regarding the pathogenesis of this rare occurrence.

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## 1. Introduction

Regional lymphadenopathy can be a diagnostic challenge. It is often caused by infections or malignancies. Among these, acute inguinal lymphadenitis is usually caused by lower extremity infection and sexually transmitted diseases, such as chancroid caused by *Haemophilus ducreyi*, lymphogranuloma venereum caused by *Chlamydia trachomatis*, genital herpes, or syphilis [1]. Other differential diagnoses include bubonic plague caused by *Yersinia pestis*, cat-scratch disease caused by *Bartonella henselae*, or tularemia caused by *Francisella tularensis*. Rarely, non-infectious diseases such as Kikuchi's disease could present as inguinal lymphadenitis [2].

*Yersinia pseudotuberculosis* is a non-spore forming, pleomorphic, non-lactose fermenting Gram negative bacillus and a member of the family Enterobacteriaceae, which is usually associated with

diarrheal illness. It also causes mesenteric lymphadenitis at the terminal ileum, which can be clinically indistinguishable from acute appendicitis (pseudoappendicitis) [3]. However, lymphadenitis in other regions caused by the organism is rare.

Herein, we report a case of a man in his 20s, who presented with unilateral inguinal lymphadenitis caused by *Y. pseudotuberculosis*.

## 2. Case report

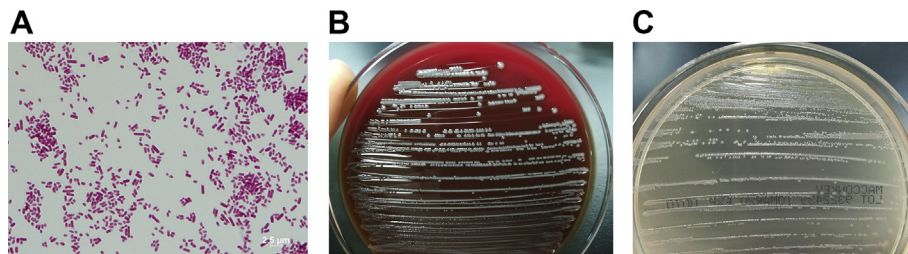
A Japanese man in his 20s without significant past medical history presented with acute onset fever and left groin pain. The symptoms started acutely 6 days prior to the presentation with left groin discomfort, which progressed to pain and fever on the following day. He denied any respiratory or gastrointestinal symptoms. He denied any sexual contact recently both with men and women. He denied any unwell contacts or animal contact. He also denied any travel abroad.

On physical examination, he was sweating and in significant discomfort. He was alert and oriented. Blood pressure was 112/78 mmHg, pulse rate was 101/minute, respiratory rate was 16/minute, and body temperature was 38.3 °C. There were multiple

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**Fig. 1.** Ultrasound imaging of left inguinal region. Multiple hypoechoic enlarged lymph nodes are seen and some were coalescent.



**Fig. 2.** Gram staining of *Yersinia pseudotuberculosis* (A) and its colonies on blood agar (B) and MacKonkey agar (C). On blood agar, the organism was cultivated for 48 hours at 35 °C with 5% CO<sub>2</sub>, and on MacKonkey agar, it was cultured aerobically for 24 hours at 35 °C.

swollen lymph nodes palpable on his left thigh below the inguinal ligament. The maximal size of the lymph nodes were approximately 3 cm. They were soft to touch and mobile with tenderness. He did not have any abnormalities in other areas, including lymph nodes at other parts of his body, joints, skin, genital, and anus. He did not have any abnormality in all four extremities including skin and nails. Rectal examination was also normal.

Laboratory tests revealed white blood cell counts of 6530 cells/ $\mu$ L with 77.6% segmented leukocytes, C-reactive protein of 4.46 mg/dL, and erythrocyte sedimentation rate of 33 mm/hour. Blood tests for

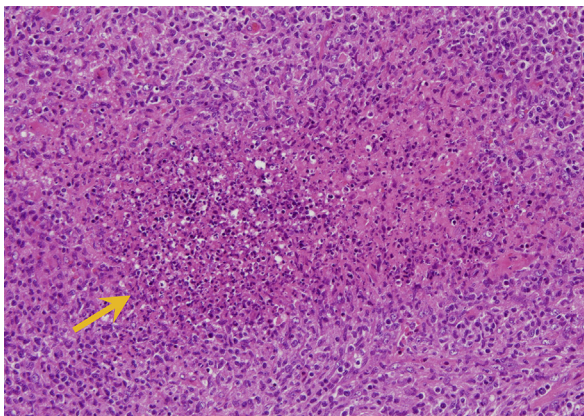
syphilis, human immunodeficiency virus (HIV) and hepatitis C were all negative but hepatitis B core antibody was positive with negative hepatitis B surface antigen/antibody. First catch urine polymerase chain reaction tests for *C. trachomatis* and *Neisseria gonorrhoeae* were also negative. Other routine blood and urine tests were unremarkable. Blood cultures were not taken.

Abdominal ultrasound test revealed multiple enlarged lymph nodes, with a maximal diameter of 31.8 mm, and some of them were coalescent (Fig. 1). There were no other abnormalities including hepatosplenomegaly.

Lymph node biopsy was performed and he received oral cephalexin for 5 days. Gram staining of the lymph node did not reveal any organisms, but the culture grew *Y. pseudotuberculosis*, which was susceptible to all antimicrobials measured (Fig. 2). He recovered from the illness in one week after the treatment. Biopsy revealed abscess formation surrounded by histocytes and lymphocytes (Fig. 3).

### 3. Discussion

Lymphadenopathy can be a diagnostic challenge. It can be acute or chronic, systemic or regional, and bilateral or unilateral. For unilateral chronic lymphadenopathy, differential diagnoses include malignancies such as lymphoma or metastases, or chronic infections such as tuberculosis [4]. For unilateral acute lymphadenopathy, it is often caused by bacterial infections manifesting as lymphadenitis [5]. Acute unilateral inguinal lymphadenitis is usually caused by sexually transmitted diseases (STD) or lower extremity infections such as cellulitis. However, our current patient denied recent sexual contact and physical examination did not



**Fig. 3.** Pathological findings using hematoxylin-eosin staining. Abscess formation (arrow) surrounded by histocytes and lymphocytes are seen.

suggest both STDs and lower extremity infections. Lymph node biopsy is often a test of diagnosis for regional lymphadenopathy and this revealed *Y. pseudotuberculosis* infection in our case. There were only a few case reports of *Y. pseudotuberculosis* infection with inguinal lymphadenopathy we could find. One reported a case of pseudoappendicitis with mesenteric lymphadenitis in 11 year old girl, accompanied by inguinal lymphadenitis [6]. The other was a case of a 6 year old Japanese boy who presented like Kawasaki disease, again accompanied by inguinal lymphadenopathy [7]. However, our case is the first to describe a case of acute unilateral inguinal lymphadenitis caused by *Y. pseudotuberculosis* as far as our literature search could find.

*Yersinia*, including *Y. pseudotuberculosis* are known to cause lymphadenitis. It is often located in the abdomen and present in the ileocecal region, and symptoms are indistinguishable from acute appendicitis. Histologically, it can present with suppurative granulomatous inflammation, and microabscesses within germinal centers [8].

We were not able to clearly identify the pathogenesis and the route of infection in this case. One hypothesis is that *Y. pseudotuberculosis* resided in the gastrointestinal tract, and lymphatic drainage at the anorectal area led the organism to reach the inguinal region, the same route as lymphogranuloma venereum caused by *C. trachomatis* but in the opposite direction [9]. Our second hypothesis is that *Y. pseudotuberculosis* was transmitted sexually by anal sex. The patient denied any recent sexual intercourse but the presence of hepatitis B core antibody could be an indication that the patient had sexually transmitted infection.

In conclusion, we had a case of acute unilateral inguinal lymphadenitis caused by a rather unusual organism. Its incidence, pathogenesis, or clinical implication should be elucidated by future studies.

## Declaration of Competing Interest

There are no conflicts of interest in this study.

## Acknowledgements

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