CASE REPORT

Yersinia pseudotuberculosis Infection Accompanied by Intussusception and Incomplete Kawasaki Disease in a 7-year-old Girl

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(Received for publication on January 25, 2021)

(Revised for publication on April 26, 2021)

(Accepted for publication on May 17, 2021)

(Published online in advance on June 8, 2021)

Infection with *Yersinia pseudotuberculosis*, a known causal pathogen of human bacterial gastroenteritis, causes various symptoms and complications. A previously healthy 7-year-old girl was admitted because of fever and gastrointestinal symptoms. She was initially diagnosed with intussusception by abdominal ultrasonography. Although the patient was successfully treated by air enema, the fever persisted. The patient was then diagnosed with incomplete Kawasaki disease based on the presence of four principal clinical features. Intravenous immunoglobulin and oral aspirin were initiated. The patient defervesced and the other symptoms subsided after the treatment. Cardiac ultrasound results showed normal coronary arteries. Because of the gastrointestinal symptoms, stool samples were cultured repeatedly, only to yield normal flora. However, serum levels of anti-*Y. pseudotuberculosis*-derived mitogen antibody were elevated between the 7th and 18th days of the disease, thereby confirming *Y. pseudotuberculosis* infection. Because *Y. pseudotuberculosis* infection results in various clinical manifestations, we must be aware of each symptom and address them systematically. (DOI: 10.2302/kjm.2021-0002-CR)

Keywords: *Yersinia pseudotuberculosis*, Kawasaki disease, intussusception, anti-*Yersinia pseudotuberculosis*-derived mitogen antibody

Introduction

Infection with *Yersinia pseudotuberculosis* (*Y. pstb*), an important causal pathogen of human bacterial gastroenteritis, causes various symptoms and complications. One severe complication is Kawasaki disease (KD). In Japan, nearly 10% of children diagnosed with KD also have *Y. pstb* infection.^{1,2} Moreover, although it is rare, intussusception is another severe complication.¹ Although stool culture is the most common method for detecting *Y. pstb*, the results are often unreliable, even when employing the cold enrichment method.³ We report a case of *Y. pstb* infection, accompanied by intussusception and incomplete KD, that was finally diagnosed using serologic testing.

Case Presentation

A previously healthy 7-year-old girl presented to our hospital during the winter period with abdominal pain and a 7-day history of fever, vomiting, and watery diarrhea. The patient denied any history of eating raw meat, drinking well water, playing in a lake or pond, animal contact, or recent travel abroad. On admission, the patient's temperature was 39.6°C, and her abdomen was flat and soft but tender to palpation. Erythema was present on the chest and both thighs. Blood tests revealed normal findings, except for an elevated white blood cell count of 19,040/ μ L and a C-reactive protein level of 16.31 mg/dL. Urinalysis also showed normal findings. Blood culture

Fig. 1 Abdominal ultrasound showed the target sign indicative of intestinal intussusception with associated mesenteric lymphadenitis.

was negative, and stool cultures revealed normal bacterial flora and the absence of causative pathogens. Abdominal ultrasound showed the target sign on the transverse colon, with associated mesenteric lymphadenitis (**Fig. 1**); consequently, intussusception was diagnosed. With the successful reduction of intussusception following air enema, the abdominal pain improved; nevertheless, the fever persisted. On the third day of admission, erythema appeared on the lips, and periungual desquamation appeared on the fingers. Cervical lymphadenopathy and conjunctivitis were not observed during the clinical course. The patient was diagnosed with incomplete KD based on the presence of four principal clinical features: persistent fever, rash, erythema on the lips, and periungual desquamation of the fingers.⁴ A treatment course including 2 g/kg intravenous immunoglobulin (IVIG) and 30 mg/kg/day oral aspirin was initiated. After the treatment, the patient defervesced, and other symptoms subsided. Cardiac ultrasound results showed normal coronary arteries during hospitalization and also at the outpatient clinic on follow-up investigations during the subsequent 3 years. Although stool samples were cultured using the cold enrichment method, no growth of *Yersinia spp.* was observed. Enzyme-linked immunosorbent assays were performed to measure serum anti-*Y. pstb*-derived mitogen (YPM) antibodies on the 7th and 18th days of the disease; the optical densities of the non-antigen-coated wells

were subtracted from those of the antigen-coated wells. Because the patient had a more than fourfold increase in serial antibody titers, a *Y. pstb* infection was confirmed. Informed consent was obtained from the patient's parents for publication of this case report.

Discussion

This is the first reported case of *Y. pstb* gastroenteritis associated with intussusception and incomplete KD. Although stool cultures did not reveal a causative pathogen, serologic examination revealed *Y. pstb* infection. The gold standard method for detecting *Y. pstb* is stool culture using the cold enrichment method. However, it is occasionally difficult to confirm the presence of *Y. pstb* through stool culture because of the low detection rate.⁵ Recent studies have reported diagnosis of *Y. pstb* infection using anti-*Yersinia* antibody measurements and loop-mediated isothermal amplification.³ In the current case, the abdominal ultrasound findings strongly suggested *Y. pstb* infection, and this was confirmed through the observation of elevated anti-YPM antibody titers. *Y. pstb* produces the exotoxin YPM, which has been shown to have superantigen properties similar to some exotoxins produced by staphylococci and streptococci.⁶

Intussusception following infections with *Y. pstb* is rare.⁷ However, it is well known that bacterial enteritis

significantly increases the risk of intussusception in children.⁸ Moreover, KD can present with intussusception, but this combination has only rarely been reported.⁹ We believe that abdominal ultrasound is a useful screening method for suspected *Yersinia* infection.¹⁰ Terminal ileitis with mesenteric lymph node enlargement is a typical presentation in *Yersinia* enterocolitis.¹¹

For patients who are immunocompetent and appear stable, antimicrobial therapy is not routinely administered in the treatment of bacterial enterocolitis. For example, a study examining the efficacy of ampicillin in those with KD and *Y. pstb* infection reported that ampicillin was not significantly beneficial in shortening the duration of clinical symptoms.¹² However, Ono et al. recommended combination therapy of IVIG and ampicillin for those with KD and *Y. pstb* infection.¹³ Our patient was older than typical KD patients, and she did not fully meet the definitive criteria of KD; however, her fever persisted even after gastrointestinal symptoms had subsided with resolution of intussusception. Consequently, we administered IVIG, without antibiotics, as treatment for incomplete KD; the patient responded fully without any further complications and the treatment was successful.

In patients with KD-like symptoms associated with intussusception, *Y. pstb* infection should be considered as a differential diagnosis, even in the presence of negative stool cultures. Because *Yersinia* infection causes various clinical manifestations, each one should be addressed individually with equal thoroughness.

Conflicts of Interest

The authors state there are no conflicts of interests.

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