CASE REPORT

Recurrent kawasaki disease with intussusception: A case report and a review of the literature

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ABSTRACT

Objective: Kawasaki disease (KD) is a multisystemic vasculitic disease that occasionally presents with gastrointestinal tract symptoms.

Case presentation: A 2-year-old girl diagnosed with KD 4 months prior was referred to our hospital with complaints of vomiting. She was subsequently diagnosed with intussusception, which was efficiently alleviated through air insufflation. However, despite her improved clinical status, she had a fever unresponsive to antibiotic therapy. Four days later, the patient exhibited typical symptoms of Kawasaki disease. Thus, intravenous immunoglobulin and high-dose aspirin were administrated, and the outcome was satisfactory.

Conclusions: In young children who present with crying, vomiting, and passage of bloody stools, color ultrasonography should be performed promptly to avoid missed diagnosis and misdiagnosis.

Key Words: Kawasaki disease, Intussusception

1. INTRODUCTION

Kawasaki disease (KD) is a form of acute autoimmune systemic vasculitis mainly affecting medium and small-size arteries. KD primarily affects the skin, mucus membrane, lymph nodes, and coronary arteries.[1] Although its etiology is still unknown, recurrent KD is considered to have an intriguing clinical manifestation. The ratio of recurrent KD cases in children varies between countries: 3%-4% in Japan, 1.9% in China, and 1.7% in the United States.[2–4] The predominant clinical problems linked to KD therapy should be preventing cardiac sequelae and inhibiting disease progression.[5,6] Several studies have examined gastrointestinal manifestations of KD, such as vomiting, diarrhea, abdominal pain, and abdominal distension.[7] However, there are limited reports on cases of KD-related intussusception.[8–11] Recurrent KD and intussusception have not been reported. This report surveys a case of recurrent KD in a child with intussusception and also examines relevant literature.

2. CASE PRESENTATION

A 2-year-old girl presented with a 3-day history of fever accompanied by vomiting, a rash was noticed one day prior to the presentation. She was admitted to the Pediatric Department of the First Affiliated Hospital of Yangtze University on July 26, 2021. A nucleic acid test for COVID-19 was negative. Her medical records revealed that she had Kawasaki disease four months prior. The patient also had a fever for more than five days and was unresponsive to antibiotic therapy. Physical examination revealed chapped lips, bayberry tongue, swollen neck lymph nodes, and bulbar conjunctival

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congestion. Therefore, intravenous immunoglobulin and oral aspirin were administered. No sign of cardiac sequelae was observed. During the 4-month interval, the child was not febrile. During the second hospitalization, red maculopapular were seen on the patient's limbs, trunk, and buttocks; some were combined, but there were no scratch marks. The patient's fingers were swollen on both hands. She also had slight bilateral bulbar conjunctival congestion. One enlarged lymph node measuring approximately 1.5 cm × 1.0 cm was palpable on both sides of the neck. A routine stool inspection revealed normal findings.

Meropenem (20 mg/kg, Q8h) was administered as an anti-infective therapy; however, this was ineffective. Therefore, we also considered the possibility that the patient may have KD. Abdominal palpation did not reveal any mass; however, color ultrasound showed a heterogeneous group in the left lower abdominal quadrant measuring 17 mm × 17 mm × 20 mm, concentric in cross-section, and sleeve in longitudinal section (see Figure 1). Therefore, she was also diagnosed with intussusception. A contrast enema was performed, and control films showed colo-colic intussusception. Air insufflation with a pressure of 60 mmHg was sufficient to resolve the intussusception. Afterward, the abdominal pain resolved, and no sign of relapse was detected on ultrasonography.

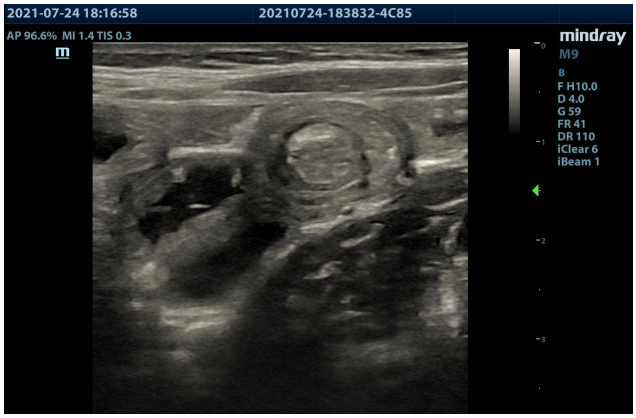

Figure 1. Color ultrasound showed a heterogeneous mass

On day 4 of admission, the child was still febrile, and antibiotic treatment appeared ineffective. Further physical examinations showed that the patient had chapped lips, strawberry tongue, and desquamation of the fingertips and anus. An echocardiogram revealed that the coronary arteries were normal. The child was diagnosed with KD, and intravenous immunoglobulins (2 g/kg), with a high-dose aspirin (50 mg/kg/day) were administered. She recovered adequately and was discharged a week later. The dose of aspirin was lowered (5 mg/kg/day) 2 weeks later till an initial normal echocardiogram. Echocardiography was repeated eight weeks later, and no sign of coronary artery abnormality was observed. Hence, aspirin was discontinued.

3. Discussion

Gastrointestinal manifestations of KD, including abdominal pain, vomiting, and diarrhea, occur in 25%-30% of cases;[12] this could lead to diagnostic delays, particularly in cases of insufficient diagnostic standards in fever. According to a former survey, 4.6% of patients with KD demonstrated acute surgical abdomen,[13] including hydrocholecystis, ischemic colitis, mesenteric and splenic ischemia, hemorrhagic duodenitis, appendicular involvement, and intussusception.

There are two possible reasons for intussusception to occur as a complication of KD. Firstly, vasculitis is an important component in KD-related pathophysiology, and recorded complications of small intestinal vasculitis among patients with the disease include jejunal stricture, hemorrhagic enteritis, and intestinal pseudo-obstruction.[14] Thus, vasculitis and submucosal hemorrhage are probably the etiological factors of Henoch-Schonlein purpura and KD.[8] Secondly, the pathogenesis for intussusception is determined by potential causes, such as lead point (Meckel's diverticulum, polyp, duplication cyst, tumor, and vascular malformation). Viral and bacterial infections can stimulate lymphatic tissues of the intestinal tract, leading to hypertrophy of the Peyer's patches at the terminal ileum, which is rich in lymphoid tissues and can function as a lead point for ileocolic intussusception.[11] These two conditions may be correlated, as KD could lead to vasculitis and aneurysm in a medium-sized submucosal vessel of the ileum or a hypertrophied Peyer's patch, which is likely to serve as a lead point for intussusception.

4. Conclusion

There are few reports of KD complicated with intussusception. However, in the treatment of KD, the possibility of intussusception should be considered if the patient is a young child presenting with symptoms such as excessive crying, vomiting, and passage of bloody stool. Furthermore, color ultrasonography should be performed in time to avoid missed diagnosis or misdiagnosis.

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Conflicts of Interest Disclosure

The authors declare they have no conflicts of interest.

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