



가와사키병에서 발생한 위십이지장 궤양 1례

손한별 · 김경민¹ · 고 흥 · 김 승

연세대학교 의과대학 소아과학교실 세브란스 어린이병원 소아소화기영양과, ¹연세대학교 의과대학 병리학교실

A case of gastroduodenal ulcer complicating Kawasaki disease

Hanbyul Sohn, Kyeongmin Kim¹, Hong Koh, Seung Kim

Pediatric Gastroenterology, Hepatology and Nutrition, Severance Children's Hospital, Yonsei University College of Medicine, Seoul; ¹Department of Pathology, Yonsei University College of Medicine, Seoul, Korea

Kawasaki disease (KD) is a systemic vasculitis associated with various clinical manifestations and complications, such as gastrointestinal abnormalities. We report a 3-year-old boy who presented with hematemesis and diffuse gastroduodenal ulcerations complicating KD. He received standard medical therapy for the disease and gastric ulcer, which showed effect after a few days. Although rare, peptic ulcers should be considered a complication of KD to ensure early diagnosis and treatment as it may cause severe morbidity.

Key words: Child; Endoscopy; Hematemesis; Mucocutaneous Lymph Node Syndrome; Peptic Ulcer

Introduction

Kawasaki disease (KD) is a systemic, idiopathic vasculitis that occurs predominantly in infants and children younger than 5 years^{1,2)}. Current paradigm regarding KD is that this condition represents an immunological reaction in genetically susceptible hosts following exposure to various environmental triggers, such as infections¹⁾. Diagnosis of KD is based on the following clinical

criteria: fever lasting > 5 days and the presence of ≥ 4 of the following 5 clinical features (oropharyngeal changes, bilateral bulbar conjunctival injection without exudate, rash, changes in the extremities, and cervical lymphadenopathy measuring ≥ 1.5 cm in diameter¹⁻³⁾). Although KD is typically self-limited, it can develop complications, including coronary artery abnormalities, ventricular dysfunction, peripheral arterial obstruction, neurological involvement, urinary abnormalities, and gastrointestinal manifestations^{1,2,4,5)}. Vomiting, diarrhea, and abdominal pain are common gastrointestinal manifestations associated with KD. Anecdotally, pseudo-obstruction, hemorrhagic duodenitis, appendicitis, and intussusception have been reported^{6,7)}.

We report a 3-year-old boy who presented with hematemesis and gastroduodenal ulcer complicating KD. To date, the gastrointestinal manifestations have not been reported as complications of KD.

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Corresponding author

Seung Kim (ORCID 0000-0003-4373-9828)

Pediatric Gastroenterology, Hepatology and Nutrition, Severance Children's Hospital, Yonsei University College of Medicine, 50-1 Yonsei-ro, Seodaemun-gu, Seoul 03722, Korea

Tel: +82-2-2228-2050 Fax: +82-2-393-9118

E-mail: pedks@yuhs.ac

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Case

A previously healthy, 3-year-old boy presented with fever and neck swelling lasting for 1 day. On the second day after being admitted to an outside hospital, he developed strawberry tongue, rash over the trunk, conjunctival injection, and edema of the hands. He received supportive therapy with antibiotics for a high index of clinical suspicion for KD. Intravenous immunoglobulin (IVIG) and aspirin were not administered due to fever lasting shorter than 5 days, making it difficult to diagnose KD. Three days after the first fever, he developed hematemesis. Esophagogastroduodenoscopy (EGD) showed gastroduodenal ulcer (Fig. 1). He was referred to Severance Children's Hospital for treatment of KD complicated by gastroduodenal ulcer.

The initial vital signs were as follows: blood pressure, 107/45 mmHg; heart rate, 157 beats per

minute; respiratory rate, 26 breaths per minute; and temperature, 39.4°C. Physical examination showed swollen neck, erythema of the tongue, oral and pharyngeal mucosa, conjunctival injection, and edematous hands and feet. No abdominal tenderness, hepatosplenomegaly or other findings suggestive of KD manifestations were noted.

Initial laboratory tests showed a hemoglobin concentration of 10.2 g/dL, white blood cell count of 6,980/mm³, platelet counts of 293,000 cells/ μ L, erythrocyte sedimentation rate of 10 mm/hr, and C-reactive protein (CRP) of 124.2 mg/L. A stool test for occult blood showed positive results. Chest and abdominal radiographs showed no abnormalities. He was diagnosed with a gastric ulcer complicating KD, and intravenous pantoprazole was administered, along with IVIG at a dose of 2 g/kg.

Compared to the previous EGD performed at the outside hospital, EGD performed on day 3 showed improvement of the ulcers. No active bleeding was observed, but diffuse, persistent ulceration was identified in the same position as before, extending along the gastric antrum to the second portion of the duodenum (Fig. 2), similar

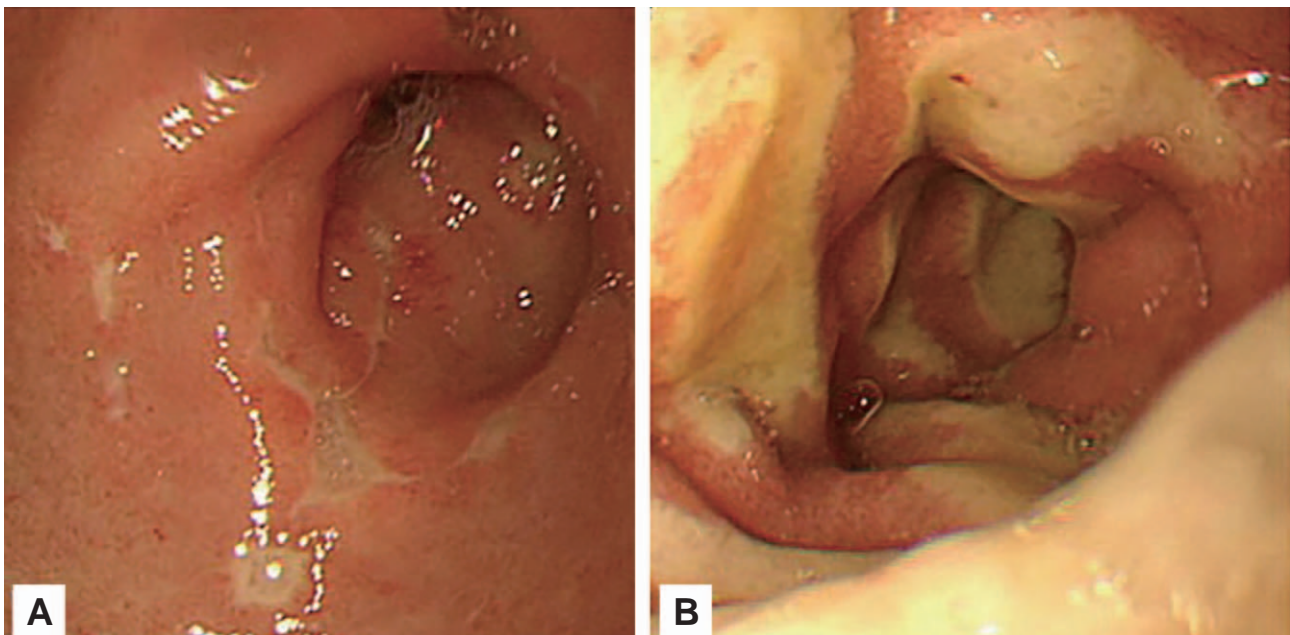


Fig. 1. Esophagogastroduodenoscopy performed at an outside hospital, showing diffuse ulceration extended along the gastric antrum (A) to the second portion of the duodenum (B).

to the EGD findings at the external hospital. Biopsy showed ulceration with acute inflammation but without any evidence of Epstein-Barr virus (EBV) or cytomegalovirus (CMV) infection (Fig. 3).

Blood and fecal cultures showed no abnormalities suggestive of bacterial infection. Polymerase chain reaction was also performed to exclude viral infections such as EBV or CMV, and the test result was negative. His serum calcitonin concentration was 1.5 pg/mL, and fasting gastrin concentrations measured on 3 separate occasions were 87.1, 19.4,

and 34.3 pg/mL, which ruled out a hypersecretory state such as Zollinger-Ellison syndrome.

High-dose aspirin was not administered owing to the ulcer, and intravenous dexamethasone therapy was initiated at a dose of 0.3 mg/kg/day to treat inflammation.

Echocardiography performed on day 8 showed small coronary artery aneurysms involving the left anterior descending artery (diameter = 2.1–2.8 mm, Z score 1.25–3.23) and the right coronary artery (diameter = 3.0 mm, Z score 3.53). Abdominal

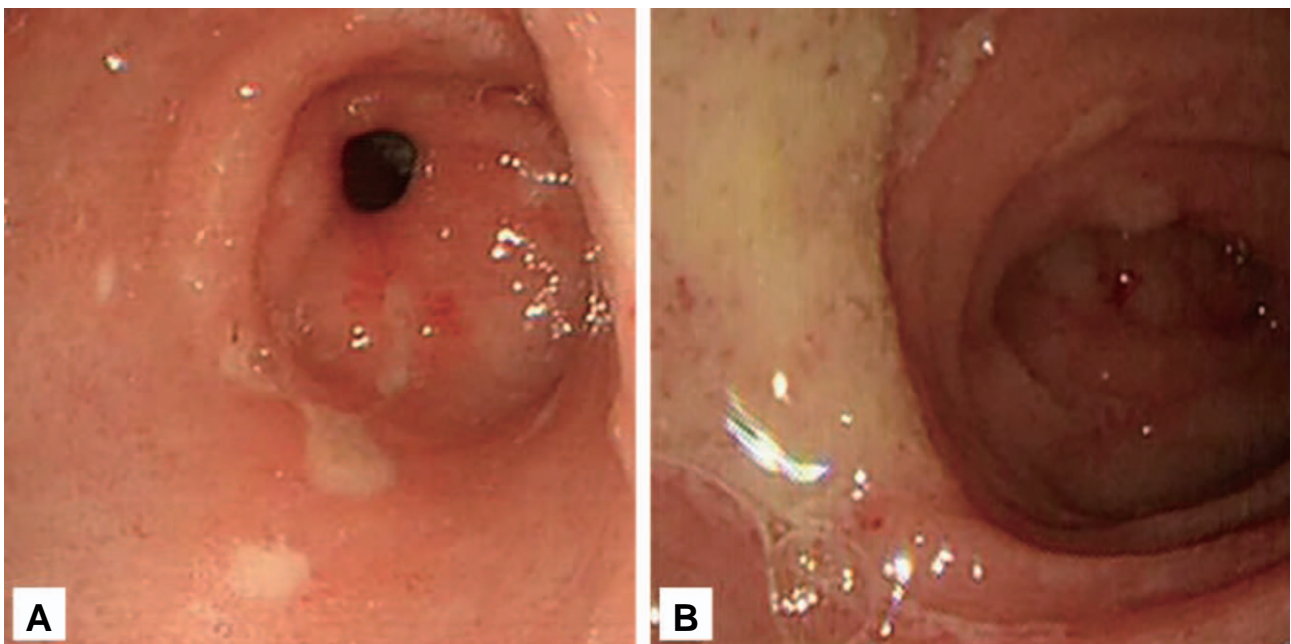


Fig. 2. Esophagogastroduodenoscopy findings on day 3, showing improvements compared with the prior findings (A, B).

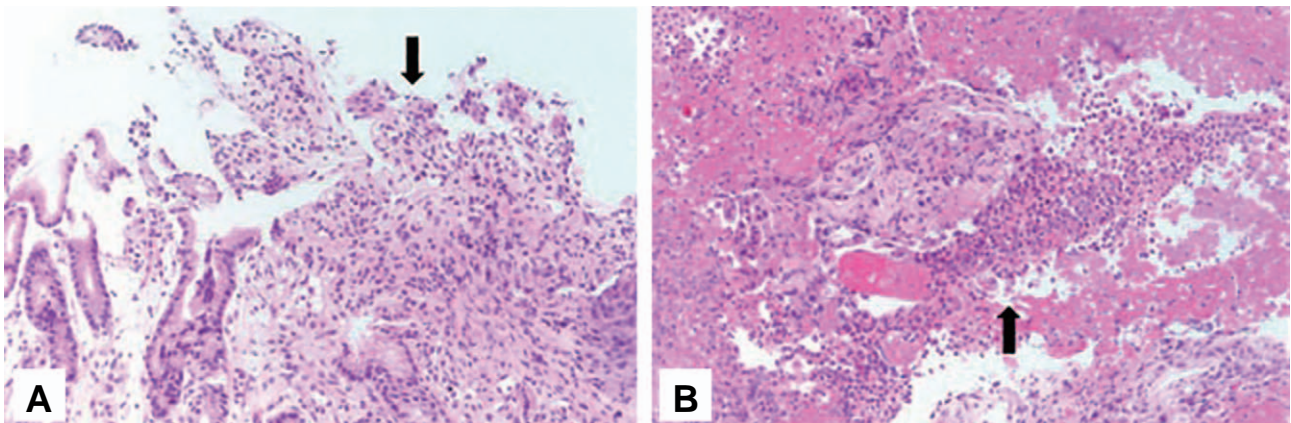


Fig. 3. Microscopic findings showing mucosal erosion (arrow) of the gastric antrum (A), necroinflammatory exudates and reactive changes of mucosal tissue (arrow) at the second portion of duodenum (B) (H & E, $\times 200$).

ultrasonography showed no abnormalities indicative of other gastrointestinal and hepatic complications. No further episodes of hematemesis occurred until day 9, and follow-up CRP level decreased to 24.2 mg/L. He was discharged, and outpatient follow-up was scheduled.

Discussion

Peptic ulcer disease is less common in children than in adults⁸. Notably, this condition can cause significant morbidity and mortality secondary to complications such as severe hemorrhage and/or perforation. *Helicobacter pylori* infection, Crohn's disease, stress, drugs, allergic or eosinophilic gastritis, hypersecretory states such as Zollinger–Ellison syndrome, and/or autoimmune diseases among other causes serve as etiopathogenetic contributors to peptic ulcer disease in children⁸.

According to reports in the available literature, several gastrointestinal complications are known to occur in patients with KD. Gastrointestinal hemorrhage is reported in patients administered high-dose aspirin after being diagnosed with KD⁹. In our patient, we observed a gastric ulcer that was not associated with treatment for KD. The ulcer in the patient was of a more severe grade and wider extent than ulcers secondary to other common causes, such as stress. Thus, the ulcer was considered a complication of KD rather than the other secondary causes. In this case, the pathological findings suggesting peptic ulcer were limited by insufficient depth of the specimens and consequent incomplete exclusion of other causes that can cause hematemesis, such as Henoch–Schonlein purpura¹⁰. The pathogenesis of peptic

ulcers in patients with KD may be attributed to persistent fever and stress secondary to vasculitis involving the stomach and duodenum. However, this theory needs more evidence.

To summarize, we reported a 3-year-old boy who presented with hematemesis and gastroduodenal ulcer complicating KD. He was treated with IVIG and dexamethasone for KD, and a proton pump inhibitor was administered to treat gastric ulcer. The latter regimen successfully treated the hematemesis, and follow-up EGD showed an improved gastric ulcer. Despite its rarity as a complication of KD, gastric ulcer may cause severe morbidity. This condition should be considered in suspicious cases to ensure early diagnosis and treatment.

Conflicts of interest

No potential conflicts of interest relevant to this article were reported.

ORCID

Hanbyul Sohn (<https://orcid.org/0000-0001-6051-0414>)

Kyeongmin Kim (<https://orcid.org/0000-0002-0454-4393>)

Hong Koh (<https://orcid.org/0000-0002-3660-7483>)

Seung Kim (<https://orcid.org/0000-0003-4373-9828>)

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References

1. Newburger JW, Takahashi M, Burns JC. Kawasaki disease. *J Am Coll Cardiol* 2016;67:1738–49.
2. McCrindle BW, Rowley AH, Newburger JW, Burns JC, Bolger AF, Gewitz M, et al. Diagnosis, treatment, and long-term management of Kawasaki disease: a scientific statement for health professionals from the American Heart Association. *Circulation* 2017;135:e927–99.
3. Brogan PA, Bose A, Burgner D, Shingadia D, Tulloh R, Michie C, et al. Kawasaki disease: an evidence based approach to diagnosis, treatment, and proposals for future

- research. *Arch Dis Child* 2002;86:286-90.
4. Tizard EJ. Complications of Kawasaki disease. *Paediatr Child Health* 2005;15:62-8.
 5. Burns JC, Shike H, Gordon JB, Malhotra A, Schoenwetter M, Kawasaki T. Sequelae of Kawasaki disease in adolescents and young adults. *J Am Coll Cardiol* 1996;28:253-7.
 6. Colomba C, La Placa S, Saporito L, Corsello G, Ciccio F, Medaglia A, et al. Intestinal involvement in Kawasaki disease. *J Pediatr* 2018;202:186-93.
 7. Zulian F, Falcini F, Zancan L, Martini G, Secchieri S, Luzzatto C, et al. Acute surgical abdomen as presenting manifestation of Kawasaki disease. *J Pediatr* 2003;142:731-5.
 8. Dohil R, Hassall E. Peptic ulcer disease in children. *Baillieres Best Pract Res Clin Gastroenterol* 2000;14:53-73.
 9. Matsubara T, Mason W, Kashani IA, Kligerman M, Burns JC. Gastrointestinal hemorrhage complicating aspirin therapy in acute Kawasaki disease. *J Pediatr* 1996;128(5 Pt 1):701-3.
 10. Chen O, Zhu XB, Ren P, Wang YB, Sun RP, Wei DE. Henoch Schonlein Purpura in children: clinical analysis of 120 cases. *Afr Health Sci* 2013;13:94-9.