Henoch-Schönlein purpura with intussusception and hematochezia in an adult

A case report

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Abstract

Rationale: Henoch-Schönlein purpura (HSP) is a common disease in children. However, HSP with intussusception and intestinal obstruction has a low morbidity in children and is occasionally seen in adults. Herein, a rare adult case of HSP complicated with intussusception and hemafecia that was successfully treated with surgery is described.

Patient concerns: A 19-year-old Chinese man suffered from HSP combined with intussusception and intestinal obstruction and presented with vomiting, diarrhea, abdominal pain, and rash.

Diagnosis: Henoch-Schönlein purpura with intussusception and intestinal obstruction.

Interventions: The patient underwent an emergency laparotomy and manual reset.

Outcome: The patient was discharged 7 days after surgery in stable condition. We followed the patient to the sixth month after surgery. This patient has no long-term complications after surgery.

Lessons: HSP with intussusception and intestinal obstruction is rarely seen in adults. Patients with HSP often present with abdominal pain as the first symptom, which is easily confused with other diseases. Once the HSP is diagnosed, surgery should be performed as soon as possible, provided there is no absolute contraindication.

Abbreviations: CT = computed tomography, HSP = Henoch-Schönlein purpura, NEUT = neutrophil count, WBC = white blood cell.

Keywords: adults, hematochezia, Henoch-Schönlein purpura, intussusception

1. Introduction

Anaphylactoid purpura was first described by Heberden in 1801. Thirty-six years later, Schönlein described the relationship between the disease and arthritis for the first time, then Henoch described the digestive tract and kidney symptoms. Thus, the disease, also called Henoch-Schönlein purpura (HSP), consists of a triad of abdominal pain, arthralgia, and purpura resulting from an immunoglobulin A-mediated small vessel vasculitis of unknown origin.^[1] Henoch-Schönlein purpura is a disease of children, with 75% of cases occurring in children between 2 and

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11 years of age. It is occasionally seen in adults. Gastrointestinal involvement is one of the most frequent symptoms of HSP in childhood (35%–65%).^[2] Major clinical manifestation of gastrointestinal involvement vary from mild abdominal pain to dark stools and hemafecia.^[3] In this paper, we reported an adult case of HSP complicated with intussusceptions and hemafecia that required surgical intervention.

Medicine

1.1. Ethical statement and consent

This study was approved by the Ethics Committee of the Second Hospital of Jilin University. The clinical and image data were obtained with the patient's consent for publication in this report. Scientific research project ethical review opinion number: (2019) Year Scientific Research Review No. (041).

1.2. Case report

A 19-year-old man suffered from right lower intermittent cramping abdominal pain after 4 hours of diarrhea. Ten days before admission, he had experienced mild vomiting and a rash on the joints and soft tissue of the extremities (Fig. 1). Laboratory studies revealed that the white blood cell (WBC) count was $13.1 \times 109/L$, Neutrophil Count (NEUT)% was 81%, and hemoglobin was 136g/L. Occult test and urinalysis result were positive, and microscopic examination found that the patient's faecal sample contained a large number of red blood cells. A computed tomography (CT) scan revealed swollen jejunal loops with the suspicious presence of a target or doughnut sign (Fig. 2). HSP with intussusception was diagnosed from typical clinical

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Figure 1. Ten days before admission, the patient had a skin rash on the joints and soft tissues of the extremities.



Figure 3. The ascending colon was completely dissociated from the posterior peritoneum and liver; the ileocecal junction intussuscepted into the ascending colon for 20 cm without perforation and necrosis.

symptoms and imaging studies. His abdominal pain continued to deteriorate. We examined the patient, and he had signs of a strong abdominal muscle tension. We diagnosed the patient with peritonitis, and an emergency laparotomy was performed. During surgery, the young man's ascending colon was completely dissociated from the posterior peritoneum and liver, and the ileocecal junction intussuscepted into the ascending colon for 20 cm without perforation and necrosis (Fig. 3). A large subserosal hematoma, as a primary cause of the intussusception, was found 15 cm from the ileocecal junction. There was no tumor or diverticulum in other parts of the small intestine and colon. Manual release of the ileocecal intussusception and ileocecal junction repair were conducted. On the first day after surgery, we gave a liquid diet after the patient's intestinal peristalsis is restored. On the third postoperative day, we gave the patient a semi-liquid diet. On the seventh day after surgery, the patient had normal food intake, and no abnormalities in exhaust and defecation. The patient was transferred to the Department of Nephrology in our hospital to continue treatment for HenochSchönlein purpura. We followed the patient to the sixth month after surgery. The patient is in good nutritional condition and the disease has not relapsed. The patient had no long-term complications after surgery.

2. Discussion

This case is the first adult patient with HSP complicated with intussusception in recent years. Henoch-Schönlein purpura, also known as an anaphylactoid purpura, is a systemic vasculitis of unknown etiology, mainly invading the skin, kidneys, joints and gastrointestinal mucosa, and serosa.^[4]

It was commonly believed that HSP was caused by an abnormal reaction to certain substances in capillaries, causing enhancement of capillary permeability, leading to extensive edema, blood clotting, and hematoma in the intestinal wall. Meanwhile, mesenteric lymph node enlargement and gastrointestinal dysfunction cause irregular intestinal wall peristalsis, resulting in an intussusception. When a segment of the intestine



Figure 2. CT scan revealed swollen jejunal loops with the suspicious presence of a target or doughnut sign. CT=computed tomography.

enters another intestine to form an intussusception, the more severe the blood circulation of the mesenteric vessels is blocked, the more easily the intestines forming the intussusception become necrotic.^[5]

HSP mainly involves the skin, kidneys, and large joints (knee, ankle, elbow, wrist, etc), causing symmetrical skin purpura and nephritis and limiting activity. The major clinical manifestation of gastrointestinal involvement includes obvious abdominal angina pain, which can be accompanied by nausea, vomiting, and diarrhea. Severe cases proceed to intussusception and perforation of the intestine. These symptoms can be easily misdiagnosed as acute gastroenteritis, peptic ulcer, necroic enteritis, acute appendicitis, and Meckel diverticulitis.^[6] These acute abdominal symptoms caused difficulties in our diagnosis. In the course of treatment, however, we performed a full abdominal CT examination on the patient in time. We combined the results of the imaging and laboratory examinations with the patient's symptoms, and we arrived at the correct diagnosis.

Ultrasonography has become one of the methods for detecting secondary intussusception in patients with allergic purpura. Under ultrasound, the cross-section of the intestine is shown as "concentric circles" and the longitudinal section shows the "sleeve" symbol. CT examination also has great diagnostic significance. It can indicate thickening of the intestinal wall edema, and can also display "concentric circles."^[7]

Different treatment strategies are employed for children and adult patients with HSP. Because HSP is a self-limiting disease, and the intestinal pressure is less than that in idiopathic intestinal obstruction, the intestinal obstruction caused by HSP can theoretically be relieved under conservative treatment.^[8] Air enema and ultrasound-guided low-pressure enema reduction can be used as the treatment of choice for children.^[9] In adult patients, intussusception often results in pathological lesions in the intestine, such as inflammation and ischemic lesions.^[10] Therefore, in older children or adults, we should operate as early as possible to avoid delays in surgery.^[10,11] In surgery for patients with shorter intussusception time and bowels in better condition, we can perform a simple manual reset. If the intestinal viability is suspicious during surgery, it is recommended that intestinal resection and intestinal anastomosis be performed to avoid iatrogenic intestinal perforation. Since HSP is a self-limiting disease, early diagnosis and avoiding unnecessary surgery are feasible. However, in the process of conservative treatment, an experienced surgical team is required to observe changes in the patient's condition closely. For patients with intussusception, the most serious complication of conservative treatment is intestinal perforation. Once there is a sign of intestinal ischemic necrosis and intestinal perforation, surgical treatment is needed.^[12,13]

3. Conclusion

In summary, patients with HSP often present with abdominal pain as the first symptom, which is easily confused with other diseases. In the process of diagnosing patients with acute abdomen pain, we should remind ourselves of the possibility of HSP as a diagnosis. Once the HSP is diagnosed, surgery should be performed as soon as possible, as long as the indications for surgery are met.

Author contributions

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References

- Diaz MC, Ramundo ML, Christopher NC. Spontaneous reduction of a small bowel intussusception in a patient with Henoch Schönlein purpura. Am J Emerg Med 2004;22:323–5.
- [2] Choong CK, Beasley SW. Intra-abdominal manifestations of Henoch-Schönlein purpura. J Paediatr Child Health 1998;34:405–9.
- [3] Chen SY, Kong MS. Gastrointestinal manifestations and complications of Henoch-Schonlein purpura. Chang Gung Med J 2004;27:175–81.
- [4] Yang YH, Yu HH, Chiang BL. The diagnosis and classification of Henoch-Schonlein purpura: an updated review. Autoimmun Rev 2014;13:355–8.
- [5] Choong CK, Kimble RM, Pease P, et al. Colo-colic intussusception in Henoch-SchoÈnlein purpura. Pediatr Surg Int 1998;4:173–4.
- [6] McCarthy R, Rosen T, Chen SH, et al. Adult Henoch-Schönlein purpura with fatal complications. Arch Dermatol 2001;137:19–21.
- [7] Shimoyama T, Matsuda N, Kurobe M, et al. Colonoscopic diagnosis and reduction of recurrent intussusception owing to Henoch-Schonlein purpura without purpura. Paediatr Int Child Health 2018;39:219–23.
- [8] Huang W-K, Fan P-C, Chen Y-C, et al. Henoch-Schönlein purpura with intussusception in an adult. Am J Med Sci 2012;344:337–9.
- [9] Golriz F, Cassady CI, Bales B, et al. Comparative safety and efficacy of balloon use in air enema reduction for pediatric intussusception. Pediatr Radiol 2018;48:1975.
- [10] Marsicovetere P, Ivatury SJ, White B, et al. Intestinal intussusception: etiology, diagnosis, and treatment. Clin Colon Rectal Surg 2017;30: 30–9.
- [11] Azar T, Berger DL. Adult intussusception. Annals Surg 1997;226:134-8.
- [12] Hetland LE, Susrud KS, Lindahl KH, et al. Henoch-Schonlein purpura: a literature review. Acta Derm Venereol 2017;97:1160–6.
- [13] Lerkvaleekul B, Treepongkaruna S, Saisawat P, et al. Henoch-Schonlein purpura from vasculitis to intestinal perforation: a case report and literature review. World J Gastroenterol 2016;22:6089–94.