CASE REPORT

Adult Intussusception Caused by *Yersinia enterocolitica* Enterocolitis

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Abstract

*Yersinia enterocolitica* (YE) infection is a rare cause of intestinal intussusception, especially in adults. We herein, report a case of adult intussusception due to YE enterocolitis. A 24-year-old woman was admitted because of severe abdominal pain. She was clinically diagnosed with ileocolic intussusception on the basis of the findings of computed tomography (CT) and a gastrografin enema. Manual surgical reduction was sufficient to alleviate the intussusception. A histologic examination of the lymph nodes around the ileocecum excluded lymphoma. Serological testing revealed that the cause of the intussusception was a YE infection. The patient’s postoperative course was good and no recurrence was seen during the follow-up.

Key words: adult, intussusception, *Yersinia enterocolitica* enterocolitis, surgery, manual reduction

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Introduction

*Yersinia enterocolitica* (YE) is a gram-negative bacillus with a worldwide distribution that grows at low temperatures (1, 2). The most common clinical manifestation of a YE infection is self-limited gastroenteritis and mesenteric adenitis. Food contamination is the most frequent source of infection in humans, usually in children (3). There are many clinical complications, but intestinal intussusception is rare (4-6). Intussusception of the bowel is defined as the telescoping of a proximal segment of the gastrointestinal tract within the lumen of the adjacent segment (7-9). This condition presents with the classic triad of cramping abdominal pain, bloody diarrhea and a palpable tender mass. Intestinal intussusception in adults is uncommon compared with the pediatric population, accounting for 5% of all intussusceptions. Bowel intussusception due to YE enterocolitis in adults is very rare (6). We herein describe a case of adult intussusception caused by YE enterocolitis, which was manually reduced under surgery.

Case Report

A 24-year-old woman presented with a complaint of right lower quadrant pain. She had no important family or medical history. The abdominal pain developed suddenly without any evident indicators, such as diarrhea or bloody stool. There was no history of overseas travel or ingestion of raw food at that time.

On examination, her temperature was 37.2°C, her heart rate was 90 beats per minute, blood pressure was 117/75 mmHg and respiratory rate was 18 breaths per minute. A fist-sized mass was palpable in her right lower quadrant, with tenderness on palpation. Her respiratory and cardiac sounds were normal, and no cardiac murmurs were audible. Laboratory tests revealed leukocytosis with elevated levels of C-reactive protein.

Computed tomography (CT) at the level of the lower abdomen revealed a target- and sausage-shaped soft-tissue mass in the right abdomen (Fig. 1A, B). The mass consisted of mesenteric vessels with a layering effect. We diagnosed the patient with ileocolic intussusception. A lead point was

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Figure 1. Abdominal CT findings. The intussusception appeared as a round target- (A, arrowheads) or sausage- (B, arrowheads) shaped mass with high density soft tissue, which represented the edematous bowel wall of the intussusciens and the intussusceptum. (C) Swelled lymph nodes were seen around the ileocecum (arrows).

Figure 2. Gastrografin enema findings. A gastrografin enema image of the colon showed a cup-shaped filling defect in the upper part of the ascending colon (A), which went up to the lower part of the ascending colon (B) during the examination. Scale bar: 1 cm

Figure 3. Intraoperative findings. The bowel wall of the reduced terminal ileum was edematous (arrowheads) and was fraught with hypertrophied lymph nodes (arrows). No necrotic changes were seen in the bowel wall.

not evident, while swelling of lymph nodes around the ileocecum was seen (Fig. 1C).

An enema examination with gastrografin, a water-soluble contrast agent, performed on the day of admission demonstrated a cup-shaped filling defect in the upper part of the ascending colon (Fig. 2A), which went up to the lower part of the ascending colon (Fig. 2B) during the examination. However, pneumatic or hydrostatic reduction of the intussusception was insufficient to restore it to its original condition. Since the severe pain persisted, surgery was subsequently performed on the same day.

The surgery revealed that the terminal ileum had telescoped into the ileocecal region (Fig. 3). The regional lymph nodes had increased in volume, and biopsy speci-
mens were sent for histological tests. No obvious lead point was palpable, and no signs of ischemia of the bowel wall were seen. Manual traction was attempted, and a 25 cm ileocolic intussusception was successfully reduced without resection. The telescoped terminal ileum appeared thickened. Histological images of the resected lymph nodes showed no neoplastic lymphoid cells and excluded malignant lymphoma (Fig. 4). There were no findings of active inflammatory responses, such as leukocyte infiltration or granuloma.

To determine the cause of the intussusception, a colonoscopy was performed on the eighth day after admission. As demonstrated in Fig. 5, aphthous erosions were seen diffusely from the terminal ileum to rectum. A microscopic examination of the terminal ileum and colon revealed slightly inflammatory responses, such as inflammatory cell infiltration, lymphoid follicles and congestion in the edematous mucosa. Non-specific acute ileocolitis was diagnosed based on these findings (Fig. 6). A stool culture did not show any specific findings. She was discharged on the eleventh day after admission.

The patient did not suffer any complications or recurrence of the intussusception during the follow-up. A serological test reported after discharge was positive for YE (serotype O3), with serum antibody titers of ×40 (7 days later) and ×160 (28 days later). An abdominal CT one month later showed mild wall thickening of the small intestine. There were no meaningful findings on CT three months later.

Discussion

YE is spread primarily via the fecal-oral route, and infection most often results from human consumption of contaminated meat products (especially pork), milk products, water and vegetables (3). Less commonly, exposure to human and zoonotic fecal carriers, including pigs, dogs, cats, and rats, results in this disease. The present patient did not eat meat products for several days before admission. Moreover, she did not have contact with any animals at that time. The source of infection was thus unknown in our case. A diagnosis is determined by testing for YE directly in the blood, feces, or in biological fluid such as ascites, or in other biological matter, or by testing for the presence of anti-YE antibodies, specifically against human pathogenic serotypes. The detection rate of stool culture is low (2, 10), and therefore, YE infection is easily overlooked. The stool culture of our patient also did not reveal YE infection. Anti-YE antibody titration is another method that can be used for making a diagnosis. Anti-YE antibodies reach a maximum concentration after 7-28 days and they remain high for about 18 months after infection (11). An increase of more than two-fold in the concentration of paired antibody titers, or more than ×160 in a single antibody titer is sufficient to make diagnosis. The serum YE O3 antibody titer in our patient was ×40 at 7 days after admission, and was increased...
many studies have suggested an association between infection and intussusception in children. Cade et al. showed an increase in the risk of intussusception in children with a history of bacterial enteritis within the previous 6 months (12). In these patients, however, no cases of intussusception followed infection with YE, perhaps due to the overall low rate of YE enteritis. In contrast, infectious colitis is not a common cause of intussusception in adults. A pathological examination of adult intussusceptions revealed that intestinal inflammatory disease was associated with only 0% to 4.5% (8, 9, 13) of all cases.

Of all 10 of the previously reported cases of intussusception associated with YE infection (Table) (4–6, 14–18), there were 9 males and 1 female. The most common locations were the ileocolic (7 cases), followed by the ileoileal (2 cases) and colocolic (1 case) region. All patients underwent surgery, and three of them required intestinal resection for signs of ischemia or sepsis. The present patient is the second case of intussusception that developed as a complication of YE enterocolitis in an adult.

In contrast to intussusceptions in children, approximately 70-90% of adults have an organic lesion that serves as a lead point within the intussusception (19-22). Many previous reports of intussusception due to YE infection suggested that the lead point was a secondary mass effect caused by lymphoid hyperplasia. In one prospective study of 261 patients (228 children and 33 adults) with intussusceptions, 88% of the cases were idiopathic, without any definitive lead point (4, 5). In these cases, the ileocecal area was the site most commonly involved (82%), with hypertrophic Peyer’s patches of the terminal ileum being responsible for 39% of the idiopathic intussusceptions of the ileocecal area. In the small intestine, an intussusception can be secondary to the presence of either intra- or extra-luminal lesions (19). Although the exact mechanism is unknown, the edematous bowel wall with thickened Peyer’s patches or the hypertrophied lymph nodes around the telescoped ileum, possibly resulting from infection, may have acted as a mechanical lead point for the intussusception in the present case.

Because of a significant risk of associated malignancy, which has been reported to be present in 65% of cases (23, 24), radiological decompression is not usually addressed preoperatively in adult intussusceptions. In fact, 70–90% of adult cases of intussusception require definite treatment, and surgical resection is generally the treatment of choice (25). However, lead points which are caused by benign diseases, such as diffuse mucosal lesions, benefit from a contrast enema evaluation, as do pediatric cases. Because CT imaging revealed no evident organic lead point in our patient, we attempted a gastrografin enema examination with
both diagnostic and therapeutic goals. Unfortunately, this procedure yielded no therapeutic results in terms of reduction. This confirmed the findings of other authors (8, 20), who reported that barium studies, despite having good diagnostic and therapeutic effects in children with a presumed diagnosis of intussusception, do not have any considerable hydrostatic reducing effect in adults.

It remains debatable whether reduction of the intussuscepting lesion should be attempted during an operation, or whether an en bloc resection should be carried out without attempting reduction (8, 25, 26). Previous reports have advocated reducing the intussusception before resection (27, 28). The potential disadvantages of this approach are intraluminal seeding and tumor dissemination via the venous flow, perforation and seeding of infection and tumor cells into the peritoneal cavity, and an increased risk of anastomotic complications (7). The advantages of intraoperative reduction, especially when the small bowel is affected, are that it may preserve a considerable length of bowel and thereby prevent the development of short-bowel syndrome. Begos and colleagues are proponents of resection without attempting reduction when the bowel is inflamed, ischemic, or friable, and in cases of obvious colocolic intussusception (with a high likelihood of malignancy) (25). In all other cases, reduction should always be attempted initially. In the present case, the intraoperative findings indicated that a large length of the small bowel was intussuscepted into the ascending colon without obvious inflammatory, ischemic, or friable changes. Therefore, manual traction was attempted in this case, and the intussusception was successfully reduced without resection.

We herein reported a case of adult intussusception due to YE enterocolitis. CT and serological testing were useful for the diagnosis. Manual reduction during surgery was sufficient to treat the patient, and she recovered with no complications or recurrence.

The authors state that they have no Conflict of Interest (COI).

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References