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Intussusception and appendicitis: What comes first?

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ABSTRACT

Intussusception and acute appendicitis are two different causes of acute abdomen that potentially require surgery. The clinical presentation can be similar in both diagnoses with symptoms including abdominal pain, vomiting, diarrhea, and fussiness but these two concern a pediatric population of different ages. We describe an uncommon CASE combining appendicular intussusception with concurrent acute appendicitis. A 27-month-old boy presented with 3 day of colicky intermittent abdominal pain, non-bilious vomit and low-grade fever. After the failure of air enema decompression, the child underwent a surgical procedure.

1. Introduction

Pediatric ileocolic intussusception is the most common cause of bowel obstruction in children under 2 years of age with an incidence of 1–4/1000 live births [1]. The etiology is often idiopathic in younger patients while in older children and adults a "leading point" is often observed [2]. The lead point may be a Meckel's diverticulum, Henoch-Schonlein purpura, lymphoma, duplication, haemangioma or an intestinal polyp. Appendix, as the lead point, has once been reported in 1923 [3]. Intussusception occurring in infancy is idiopathic, i.e. without any lead point, in approximately 90% of cases. Common symptoms are intermittent colicky abdominal pain and distension, palpable mass, bilious vomiting and rectal bleeding. Most cases can be managed conservatively by hydrostatic or pneumatic reduction while only a few children require laparoscopic or open reduction with/without bowel resection.

Acute appendicitis is the most common cause of urgent abdominal surgery in children with a peak incidence of 25/10,000 between 10 and 17 years of age while it is uncommon between 1 and 4 years of age. However, coexisting intussusception and acute appendicitis has been rarely described. The more frequently reported association has been appendix entrapped in the ileocolic intussuscepted segment [4,5].

We report on a rare clinical case of intussusception and appendicitis on a pre-scholar child with review of literature. We also discuss the clinical consequences that invagination of appendix may cause failure of pneumatic reduction of intussusception and the need for surgery.

2. Case report

A 27-month-old boy was admitted for colicky intermittent abdominal pain, non-bilious vomit and low-grade fever. Symptoms started 3 days earlier but overall clinical conditions were discrete. At physical examination, the abdomen was distended with defense in the right and left lower quadrants. Laboratory investigation revealed only a mild increase in *C*-reactive protein with normal WBC and liver function tests. At abdominal ultrasound, a clear finding of ileocolic intussusception was observed in the right hypogastrium with typical target sign Mild fluid collection and reactive lymph nodes were seen around the invaginated segment. Barium enema confirmed the diagnosis of ileocolic intussusception at the level of the transverse colon. Under anaesthesia, hydrostatic reduction was attempted with incomplete reduction of invaginated segment. Then, we decided to proceed with surgery. At laparoscopy, a free inflamed hyperemic appendix was observed between the ileal loops (Fig. 1). The terminal ileum was still invaginated for 10 cm into the ascending colon. Laparoscopic reduction of the invaginated ileal loop was easily performed. Several adhesions from mesentery of terminal ileum to ileocecal valve were observed and then resected. At the same time, appendectomy was also removed. Histopathological report described an appendix with a congested serosa and a bluish mucosa without sign of bacterial infection. The diagnosis was subacute or chronic appendicitis with hemorrhagic findings. The child started enteral feeding on the following day and was discharged on the 4thpost-operative day.

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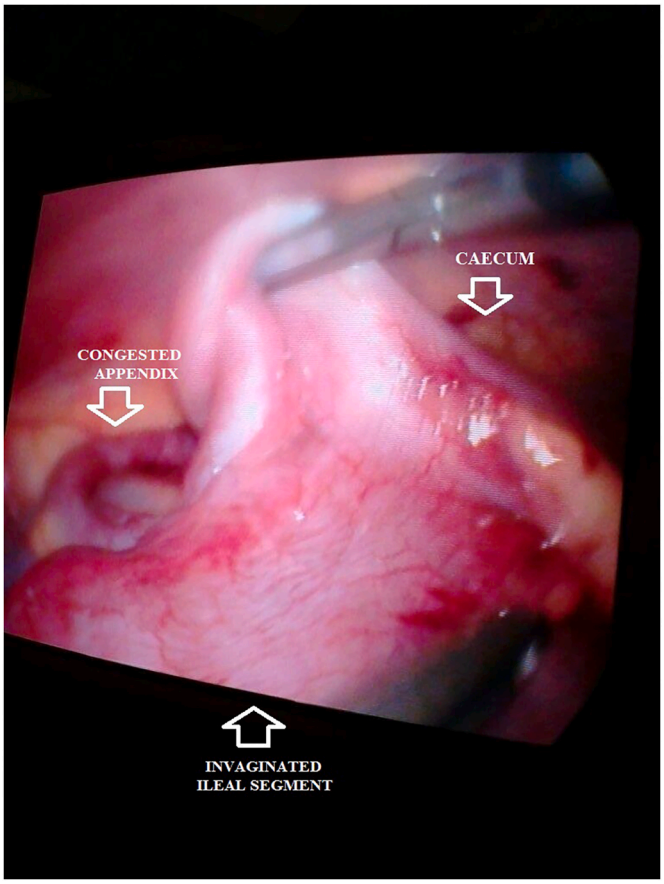

Fig. 1. At laparoscopy, a free inflamed hyperemic appendix was observed between the ileal loops.

3. Discussion

We reported an unusual case of intussusception apparently associated with primary acute appendicitis. Actually, at the laparoscopy, we found an apparently inflamed appendix between the normal ileal loops and then we proceeded with an easy reduction of terminal ileum, which appeared congested but viable. However, the histopathological examination showed findings not consistent with acute appendicitis but wall changes suggestive of vascular congestion (intraparietal hemorrhages, congested serosa and bluish mucosa). Inflammatory infiltrates, typical of acute appendicitis, were not observed. On this basis, we concluded that the appendix was initially entrapped in the invaginated segment. This may have caused the vascular changes on the appendix wall and lumen. Most likely, partial reduction following contrast enema has released the appendix, which became free in the abdomen.

In literature, few cases of coexistent intussusceptions and appendicitis have been reported, especially in children. Thomson reported on a 22-month-old baby affected by caecum-colic intussusception with acute appendicitis and peritonitis. He found perforation at appendix basis while its distal portion was only congested. The apex of intussusceptions was the caecum at the level of appendix basis. He concluded that the presence of a perforated gangrenous segment of the appendix preceded intussusception that otherwise would not have caused peritonitis [6]. Bevan PG reported a case of a 5-year-old girl who underwent open reduction for intussusception. He found an apparently innocent slightly swollen appendix which, when opened, contained pus with ulcerated mucosa. He concluded that the first event was intussusception and that acute appendicitis was caused by the occlusion due to intussusception [3]. More recently, Min Kee reported a case of a 38-month-old boy with suspected acute appendicitis who underwent CT scan. It showed an ileocolic intussusception with trapped appendix within the intussusception. At surgery, they found an inflamed appendix within the intussusception, which was easily reduced. Histological examination of appendix showed only early inflammatory infiltration. They concluded that patients with intussusception should be suspected to have associated diseases such as acute appendicitis [7]. Marjon L et al. has recently reported on a 1-year old infant who was referred with suspected intussusception. At ultrasound the appendix was not evident and a partial reduction was obtained with enema. At laparoscopy, the appendix resulted within the intussusception and inflamed. However, they not address if the intussusception preceded the appendicitis or the other way around [8]. Betancourth-Alvarenga JE reported on three children <14 years old who underwent surgery for acute abdominal pain and who had coexistent invaginated appendix within the intussusception [9]. Interestingly, in all these reports as well as in our clinical situation, the appendix was always trapped or suspected to be involved in the intussusception process.

The etiology of acute appendicitis is still poorly understood. However, invasion of the appendiceal wall by bacteria seems to be the ultimate event. Currently, obstruction of appendix lumen, from whatever causes, is considered the major factor in the pathogenesis of acute appendicitis. On this basis, we believe that acute appendicitis observed in cases of intussusceptions is always a secondary event to obstruction of appendiceal lumen. In fact, none of the reported cases had an inflamed acute appendix that was isolated and not involved in the intussusceptions process. In fact, the involvement of the appendix in intussusceptions is an extremely rare clinically evident condition while in the old surgical era it was observed more frequently. In fact, Bevan GP stated that in 12 cases of surgically treated intussusceptions, the appendix often remained blue or showed incipient gangrene, so he performed appendectomy in five of them [3]. Recently, Wy Wong et al. reported on 173 cases of pediatric intussusception over a 17-year period: 160/173 underwent a pneumatic or hydrostatic reduction with a success rate of 79.4% [10]. In this extensive experience no cases of intussusception and acute appendicitis have been mentioned [11]. More recently, Chan TP et al. reported a case on a 30-month-old child suffering from intussusception and successfully treated with a contrast air enema which, in the following days, showed a clinical worsening leading to a surgical exploration. The authors found perforated appendicitis with appendicitis involved in intussusception [11]. Wan Yee T et al. reported a similar case of easily resolved intussusception with air reduction, which subsequently presented with perforated appendicitis [12]. Marjon L et al. reported on a 1 year old child presented with intussusception unresponsive to pneumatic reduction. At surgery they found the appendix within the invaginated segment clearly inflamed and, then, removed [8]. Today, the management of intussusceptions is often non-surgical and associated with antibiotics treatment. These two important advancements may have played a major role in the rare observation of this condition and in the prevention of acute appendicitis following non-surgical reduction in current clinical settings. However, we should keep in mind that appendix may be part of intussusception and which may further complicate the clinical course of those children with the need of surgery.

In conclusion, we believe that incidental acute appendicitis in children affected by ileo-colic intussusception should be considered as a further complication of the intussusception event. Furthermore, a few days of antibiotics following reduction of intussusceptions should be administered independently from the clinical outcome to prevent the risk of subsequent acute appendicitis. In fact, this complication may cause a more severe evolution of ileo-colic intussusception which may require surgery or even bowel resection.

Patient Consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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Authorship

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Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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