Case Report

Recurrent ileocolic intussusception with appendiceal lead point in a 2-year-old child: an etiology to be aware of on ultrasound✩,✩✩

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Abstract
Ileocolic intussusception is a differential consideration in young pediatric patients presenting with acute abdominal pain. Appendiceal intussusception is an uncommon variant of ileocolic intussusception where the appendix is contained within the intussusception, which can be challenging to diagnose preoperatively. In this case report, we present a 25-month-old female presenting with intermittent, diffuse abdominal pain. Initial ultrasound evaluation demonstrated ileocolic intussusception, which was successfully reduced by air enema. The patient experienced recurrent symptoms and had several recurrent episodes of ileocolic intussusception with the appendix contained within the intussusception. After the fifth recurrence, the patient underwent surgical intervention. The inflamed appendix was discovered to be inverted within the cecum, resulting in a lead point for intussusception of the terminal ileum. The patient underwent laparoscopic reduction of the ileocolic intussusception and appendectomy. Few cases of appendiceal and concurrent ileocolic intussusception are described in the literature. It is important that radiologists and surgeons be aware of this entity when evaluating pediatric ileocolic intussusception, particularly at the time of ultrasound and air enema, in order to ensure appropriate management and prevent complications.

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Introduction

Appendiceal intussusception was first described in 1858 by M’Kidd, MRCS [1]. It is considered a rare entity, where the appendix is inverted into the cecum [2]. It is difficult to diagnose preoperatively [1,3]. Based on a study of 71,000 human appendix surgical specimens, the overall incidence is approximately 0.01% [4]. Appendiceal intussusception, in rare instances, can act as a lead point for secondary ileocolic intussusception [5]. Presenting symptoms typically include intermittent, crampy abdominal pain. Clinically, it can mimic more common acute pediatric conditions, such as appendicitis [3]. For pediatric patients presenting with acute abdominal pain, radiologists should be aware that ileocolic intussusception with appendiceal intussusception may be a differential consideration to help guide management and prevent potential complications.

Case description

A 25-month-old female with a history of constipation presented with intermittent, generalized abdominal pain for 2 weeks duration. Associated symptoms included fussiness, anorexia, and lethargy during painful episodes. There were no constitutional symptoms such as fever or recent illness. The patient had no additional gastrointestinal symptoms such as diarrhea, vomiting, or a pattern of abnormal bowel movements. She was treated by her Pediatrician with Miralax 1 week prior to presentation, though this did not alleviate her painful episodes. On presentation to the emergency department, physical examination was normal, including a benign abdomen and normal hemodynamics. No laboratory studies were performed. Initial ultrasound demonstrated ileocolic intussusception. There was no pneumoperitoneum on an abdominal radiograph. The patient underwent successful ileocolic reduction by fluoroscopic air enema. The following day, the patient had 2 recurrent episodes of abdominal pain. She had 2 subsequent ultrasounds demonstrating ileocolic intussusception, and underwent 2 additional successful uncomplicated air enema reductions. At the time of second reduction, a sausage-shaped density was identified in the right lower quadrant, which was hypothesized to represent an inflamed appendix and edematous mesenteric fat (Fig. 1). The following day, the patient’s pain recurred, and ultrasound demonstrated an intussusception comprised of inverted appendix, mesenteric fat, and terminal ileum within the intussusception, as well as mural thickening of the cecum intussusceptum (Fig. 2). The patient once again underwent successful air reduction enema. This cycle of recurrence and reduction continued, and after the fifth recurrence of appendiceal and ileocolic intussusception, the patient underwent laparoscopic reduction as well as laparoscopic appendectomy. Intraoperative findings included an inflamed and inverted appendix acting as the lead point for intussusception of the terminal ileum (Fig. 3). All bowel appeared viable at the time of intervention. The appendix was inflamed without signs of perforation. Pathology demonstrated appendiceal inflammation with diffuse mucosal ulceration, measuring 8 mm in average diameter and 5 cm in length, and containing hemorrhagic purulent fecal material. The patient’s postoperative course was uncomplicated and she was discharged on postoperative day one.

Discussion

Ileocolic intussusception usually occurs in children between the ages of 6 months and 2 years, with classic symptoms including acute, intermittent abdominal pain, currant jelly stools, a palpable abdominal mass, vomiting, diarrhea, or fever [6,7]. Most cases are idiopathic, with no abnormality demonstrated except hypertrophied lymphoid tissue [6]. Only 5% of cases have an identifiable lead point such as Meckel diverticulum, duplication cyst, tumor, or polyp [6]. In our case, the lead point was identified to be an inverted, inflamed appendix with
highly imaging appendix, intussusception reductions, lena. Pons demonstrated recognition, suggesting intussusception [8]. Population secondary to identify cecum. Mesenteric inflammation in appendix [5].

Fig. 2 – Gray-scale ultrasound images of the right lower quadrant ileocolic intussusception. The appendix (A) is demonstrated in its long axis within the intussusceptum (white arrow), as well as mesenteric fat (B) (white arrow) as is the terminal ileum in its short axis (C) (white arrow).

Fig. 3 – Intraoperative photographs identify the base of the intussuscepted appendix (A) (white arrow) and the inflamed appendix after reduction (B) (white arrow), prior to appendectomy.

mesenteric fat, pulling the terminal ileum into the intussusceptum.

Appendiceal intussusception is an uncommon but established entity that was first described in 1858. It is rare to identify appendiceal intussusception as an implied lead point for secondary ileocolic intussusception [5]. The average age of presentation for appendiceal intussusception in the pediatric population is 16 years of age, with a slight male predilection [8]. Most reported patients presenting with appendiceal intussusception had chronic intermittent symptoms (63%), waxing and waning over weeks to months [9]. One study demonstrated multiple presentations for appendiceal intussusception, with symptoms divided into 4 groups. The first group demonstrated an onset similar to acute appendicitis. The second group demonstrated symptoms consistent with intussusception. The third group had a prolonged presentation with intermittent attacks of pain with or without vomiting and melena. The fourth group demonstrated no symptoms [5].

Appendiceal intussusception is important to recognize, as it can present as a filling defect within the cecum on enema reductions, and can be mistaken for a cecal mass. Appendiceal intussusception can occur spontaneously without underlying abnormality, but anatomic variations such as a mobile appendix or cecum can be contributory [2]. Ultrasound is the imaging modality of choice to diagnose intussusception; it is highly sensitive and specific. In the transverse plane, the alternating concentric hypoechoic and echogenic layers have a signature “target” or “donut” sign appearance [6].

In the absence of contraindications such as pneumoperitoneum or peritonitis, the initial treatment of ileocolic intussusception is reduction by fluoroscopic air enema [6]. However, recurrences are not uncommon [6,10]. The definitive treatment of appendiceal intussusception is intraoperative reduction and appendectomy [2]. The appendix is often inflamed at resection [9].

In this report, we described a case of a 25-month-old child with appendiceal intussusception treated initially with fluoroscopic air contrast enemas and subsequently with laparoscopic reduction and appendectomy. The appendix is important to consider in children as a potential lead point for intussusception, and this diagnosis should be considered at the time of ultrasound and air enema to help guide management and prevent complications.

Patient consent

Patient consent obtained for the publication.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2022.03.040.

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