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Case Report

A very rare case of ileocolic and appendiceal intussusception with acute appendicitis ☆☆☆

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ABSTRACT

Intussusception is a common cause of bowel obstruction in children, typically occurring in those under 3 years old and often idiopathic. Secondary intussusception is less common in pediatric patients and usually involves a pathological lead point. Appendiceal intussusception is rare, occurring in only 0.01% of appendectomy specimens, and can mimic other acute abdominal conditions, making preoperative diagnosis challenging. We report a case of a 7-year-old male who presented with a 3-day history of crampy right lower quadrant pain and nonbilious vomiting. Ultrasound revealed an ileocolic intussusception with a suspected pathological lead point. Further imaging identified a distended, non-compressible appendix within the intussusceptum, leading to a diagnosis of secondary ileocolic intussusception due to acute appendicitis. Hydrostatic reduction was initially successful, but the patient developed recurrent intussusception within 24 hours. Surgical exploration confirmed McSwain type 3 appendiceal intussusception, necessitating manual reduction and appendectomy. The patient recovered well postoperatively. Clinicians should maintain a high index of suspicion for appendiceal pathology in older children with intussusception, and thorough imaging evaluation is essential. Early recognition of an inflamed appendix as the lead point is critical to prompt appropriate surgical intervention, ultimately preventing further complications. This case contributes to clinical practice by emphasizing the need for tailored diagnostic and therapeutic strategies in managing rare, complex presentations of intussusception.

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Introduction

Intussusception is one of the most common causes of acute abdominal pain in children. It occurs mostly in those under 3 years of age, with the highest incidence between 5 and 10 months old [1]. It occurs when a segment of the bowel (the intussusceptum) telescopes into an adjacent segment (the intussusciptum), leading to bowel obstruction, ischemia, and potential perforation if left untreated [2]. The condition is idiopathic in approximately 90% of pediatric cases, often attributed to lymphoid hyperplasia in the terminal ileum acting as a lead point [3]. However, in older children and adults, secondary intussusception is more common and is usually caused by a pathological lead point. Common lead points include Meckel's diverticulum, polyps, and lymphoid hyperplasia. Rarely, appendiceal pathology can also act as a lead point [4].

Appendiceal intussusception is an exceptionally uncommon condition, with an estimated incidence of 0.01% among patients undergoing appendectomy [5]. It involves the appendix invaginating into the cecum, which can mimic various acute and chronic abdominal disorders, rendering preoperative diagnosis challenging [6,7].

Here, we present a case of ileocolic and appendiceal intussusception in a child, with acute appendicitis as the pathological lead point.

Case presentation

A 7-year-old male presented with a 3-day history of crampy right lower quadrant abdominal pain and nonbilious vomiting. Over the first 2 days, his pain was intermittent and relatively mild, but it progressively worsened by the third day, prompting his family to seek medical care. On arrival, his vital signs were as follows: temperature 37.5°C, heart rate 110 beats per minute, respiratory rate 20 breaths per minute, and blood pressure 102/68 mmHg. Physical examination revealed localized tenderness in the right lower quadrant.

Laboratory tests showed a normal white blood cell count (WBC) with a left shift. C-reactive protein (CRP) was mildly elevated at 12 mg/L, while procalcitonin was unavailable. An abdominal ultrasound performed on presentation identified an ileocolic intussusception in the subhepatic region, characterized by a target sign (Fig. 1). Due to the patient's atypical age for idiopathic intussusception, a lead point was suspected. Further imaging revealed a distended, noncompressible appendix invaginating into the cecum as part of the intussusceptum, alongside the terminal ileum (Figs. 2 A and B). This led to a diagnosis of secondary ileocolic intussusception with acute appendicitis as the lead point.

Later that same day, the surgical team initially attributed appendiceal edema to entrapment within the intussusception and opted for hydrostatic reduction, which was successful. A postreduction ultrasound demonstrated persistent dilatation and noncompressibility of the appendix, but the patient showed no immediate complications at that time.

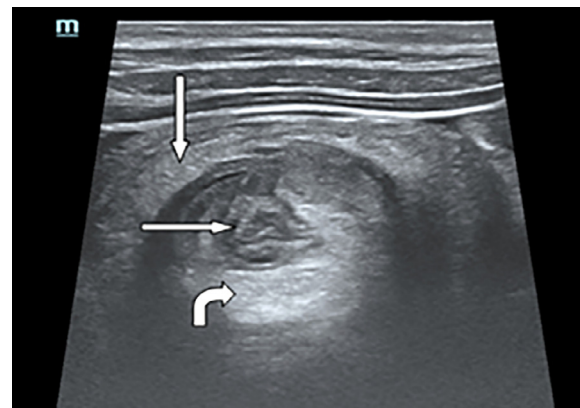


Fig. 1 – RUQ transverse scan shows the terminal ileum (horizontal arrow) and echogenic mesenteric fat (curved arrow) telescoping into an edematous, thickened cecum (vertical arrow), appearing as a target sign.

The following day, the patient returned with recurrent abdominal pain, and repeat ultrasound confirmed recurrent ileocolic intussusception, again involving the inflamed appendix. Surgical exploration was undertaken, revealing both an inflamed, distended appendix and ileocolic intussusception. The appendix was found invaginating into the cecum, consistent with McSwain type 3 appendiceal intussusception (Fig. 3). Manual reduction of both the ileocolic and appendiceal intussusceptions was performed, followed by appendectomy. The patient recovered well postoperatively.

Discussion

Appendiceal intussusception occurs when the appendix invaginates into the cecum. It is most commonly associated with appendiceal tumors, inflammation, or idiopathic causes [8]. Its pathophysiology is not fully understood but is thought to result from abnormal motility or inflammation leading to invagination [9].

The McSwain classification describes appendiceal intussusception in 5 types based on the degree and pattern of invagination. In Type 1, the appendix folds into itself, creating a “clock spring” appearance. Type 2 involves the appendix invaginating into the cecum with the tip remaining free. In Type 3, the appendix (including its base) invaginates into the cecum without inversion of the cecal wall, as observed in our case. Type 4 occurs when the appendix and cecal wall invaginate together into the colon, while Type 5 involves the appendix and ileum invaginating together into the cecum [4,10].

In many Type 3 cases, a localized appendectomy is sufficient to remove the lead point and prevent complications such as recurrent intussusception, ischemia, or necrosis, provided the cecal wall is not involved [7,11]. By contrast, when the inversion extends into the cecal wall (Types 4 and 5), more extensive bowel resection may be required to remove all compromised tissue [7,11]. Intraoperative recognition of the Mc-

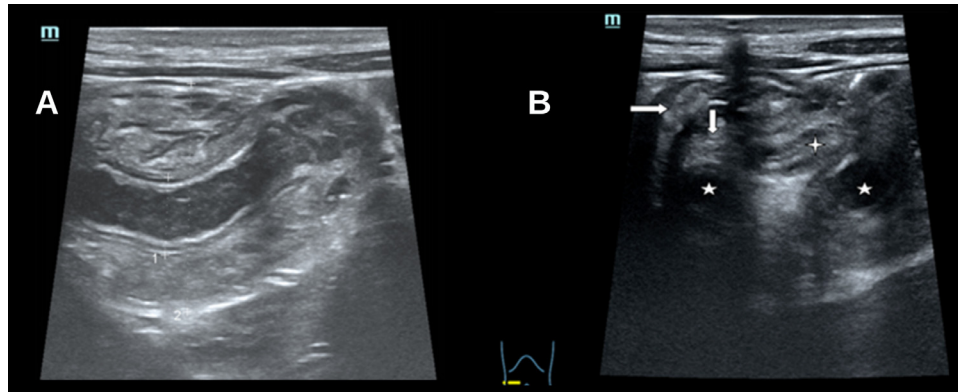


Fig. 2 – (A) RUQ longitudinal scan shows a distended, noncompressible appendix invaginating into the cecum as part of the intussusceptum, alongside the terminal ileum. **(B)** Transverse scan shows the intussusciens as a thickened cecum (horizontal arrow) and the intussusceptum as a distended appendix (stars), echogenic mesenteric fat (vertical arrow), and terminal ileum (cross).

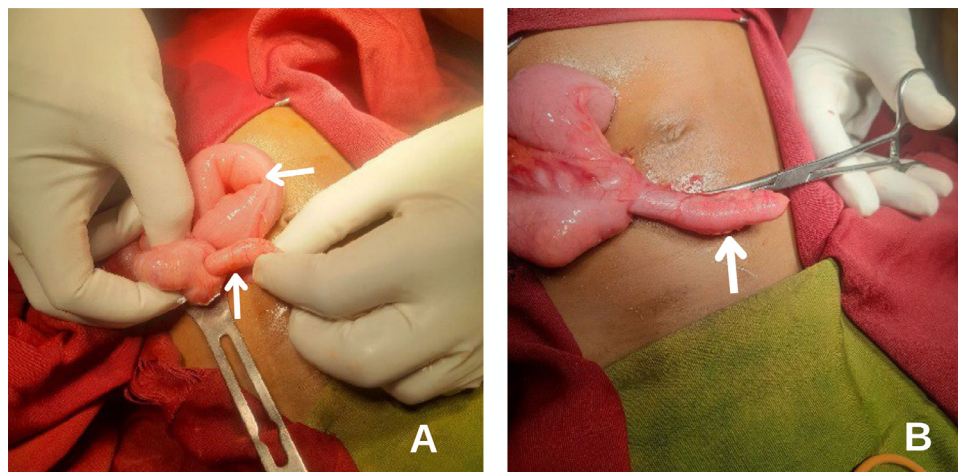


Fig. 3 – (A) Intraoperative image demonstrating a distended appendix (vertical arrow), and **(B)** distended erythematous appendix after manual reduction.

Swain classification guides surgeons in selecting an approach that minimizes unnecessary resection while adequately addressing the pathology [7,11,12].

Management of appendiceal intussusception varies depending on severity and response to initial treatment [13–17]. Some reports describe successful initial attempts at non-operative reduction using fluoroscopic air enema or hydrostatic reduction, particularly when ileocolic intussusception is present. However, these approaches often fail when the appendix acts as a lead point or is inflamed, leading to recurrence [14–16,18,19]. Previous studies suggest that surgical intervention is required if reduction is unsuccessful, intussusception recurs, or appendicitis is suspected. In several cases, laparoscopic reduction and appendectomy were performed successfully [14–16,20], while laparotomy was favored in cases with extensive bowel involvement, inflammation, or gangrenous changes [13,17].

Our case differs from previously reported cases in several key aspects, including patient age, diagnostic imaging find-

ings, management approach, and recurrence pattern. Most reported cases of appendiceal intussusception with ileocolic intussusception occur in much younger children, typically under 3 years old, such as in the cases described by Kee et al. (38-month-old), Lee et al. (35-month-old), and Furlong et al. (25-month-old), making our case of a 7-year-old child a rare presentation [8,9,14]. Additionally, unlike the cases by Ebrahimi and Yeh, where the diagnosis was only confirmed intraoperatively, our case stands out for its clear preoperative ultrasound identification of a distended, noncompressible appendix within the intussusceptum, which allowed for early recognition of the pathological lead point [21]. The management approach in our case included an initial attempt at hydrostatic reduction, which was successful. However, the patient experienced recurrence within 24 hours, requiring surgical intervention. Hydrostatic reduction was initially chosen because it is the first-line treatment for ileocolic intussusception in stable patients and avoids the risks of surgery [19]. Nevertheless, surgical intervention remains the definitive treat-

ment when a lead point is present, as nonoperative reduction often fails or leads to recurrence, as seen in this case [14,20]. This is similar to the case by Furlong et al., where multiple recurrences led to eventual appendectomy, but differs from the cases by Kee et al. and Lee et al., where surgery was performed immediately after failure of pneumatic reduction [8,9,14]. In contrast to Algu's case, which required a right hemicolectomy due to ischemia, our patient underwent manual reduction and appendectomy without bowel resection, reinforcing that early diagnosis can prevent extensive surgical intervention [17].

Conclusion

Atypical lead points, such as appendiceal pathology, should be considered in older children presenting with intussusception. Clues suggesting appendiceal involvement include an atypical age, recurrent intussusception after successful reduction, and ultrasound findings of a distended, noncompressible appendix within the intussusceptum. Early recognition through imaging and awareness of these clues can guide appropriate management, preventing recurrence and complications. This case reinforces the importance of preoperative ultrasound in identifying appendiceal pathology as a lead point and highlights the need for a high index of suspicion among pediatricians, radiologists, and surgeons when evaluating intussusception in older children.

Patient consent

Written informed consent was obtained from the patient's parents for publication.

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