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An Extremely Rare Coexistence: Acute Appendicitis and Multiple Intussusceptions in an Adult

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Background:	Adult intussusception is a rare phenomenon, acute appendicitis accompanying multiple transient intussusceptions are much more uncommon. Satisfaction and quitting imaging studies after finding an intussusception on ultrasound, may lead diagnostic errors. Radiologists should raise their awareness of imaging findings in intussusception and keep in their mind coexistent troubles in the belly. This unique case presents unusual imaging findings of a rare dual abdominal emergency condition, particularly highlighting the value of abdominal computed tomography.
Case Report:	32-year-old female was admitted to Emergency Department with complaints of epigastric abdominal pain and vomiting. US identified 'target' appearance on left paramedian location at umbilical level. Contrast enhanced abdominal CT not only confirmed the enteric intussusception that was demonstrated on previous US, but also showed additional concomitant intussusceptions and inflamed appendix.
Conclusions:	Adult intussusception is a rare phenomenon, multiple transient intussusceptions are even more uncommon. This unique report adds, precious clinical and imaging findings of acute appendicitis coexisting with multiple spontaneously resolving intussusceptions, to the literature. Physicians should be alerted for accompanying multiple abdominal pathologies and use justification essentials to make their decisions about the selection of the appropriate imaging modality.
MeSH Keywords:	Appendicitis • Intussusception • Multidetector Computed Tomography • Ultrasonography
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Background

Acute appendicitis is the most common acute nontraumatic surgical abdominal emergency both in pediatric and adult patients [1]. Intussusception is much more prevalent in pediatric populations and is a rare entity in adults, representing 5% of all intussusceptions [2]. Both of them have different etiologies. Moreover, clinical signs and symptoms, physical examination, and laboratory findings can be non-specific. Imaging may be helpful but clinical presentations should be evaluated carefully. Since what you may find is what you are looking for, physicians in emergency departments as well as surgeons and radiologists, should be ready to investigate all the signs and findings that may guide them to a correct diagnosis. The satisfaction of success may be dangerous if a stomach has more than one acute abdominal diseases. We present an adult patient presenting with acute abdominal pain, in whom computed tomography (CT)

revealed acute appendicitis, in addition to three enteric intussusceptions, that one of them was discovered on the previous ultrasound (US). Surgery confirmed appendicitis but intussusceptions were found to be resolved spontaneously. To our knowledge, this is a unique case of a dual abdominal emergency condition investigated with imaging, ever reported in the English literature.

Case Report

A 32-year-old female was admitted to the Emergency Department with complaint of epigastric abdominal pain, without umbilical presentation or migration to any direction. No history of previous abdominal surgery was declared by her. She reported two episodes of vomiting before admission. Laboratory findings were inconclusive, apart from the increased white blood count of 12500/mm³ with polymorphonuclear leukocyte dominance. Physical

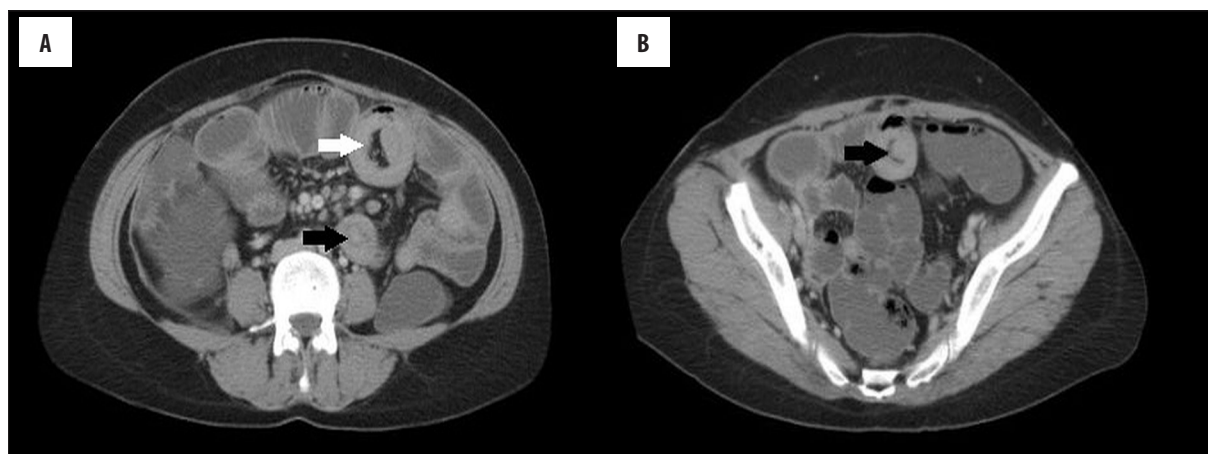


Figure 1. Contrast material-enhanced CT scan of the abdomen demonstrates the 'bowel within bowel' appearance representing the intussusception, which was discovered on previous US (white arrow in A). Additionally two accompanying intussusceptions are readily identified (black arrow in A, black arrow in B).



Figure 2. Contrast material-enhanced CT scan demonstrates enlarged and enhancing retrocaecal appendix with periappendiceal fat stranding (arrow).

examination revealed slight abdominal wall defense and rebound pain with increased bowel sounds on auscultation. Neither costovertebral angle tenderness nor urinary abnormalities on urine analysis were detected. Abdominal radiograph on erect position did not show air fluid level or free subdiaphragmatic intraabdominal air. US identified 'target' appearance on left paramedian location at umbilical level. Contrast-enhanced abdominal CT not only confirmed the enteric intussusception that was demonstrated on previous US, but also showed two additional concomitant intussusceptions (Figure 1A, 1B) and inflamed appendix located behind the gas-filled caecum and ascending colon. The appendix was enlarged, thicker than 10 mm in diameter, showing wall thickening and abnormal enhancement with fat stranding around it (Figure 2). No signs of perforation or plastron formation were found. Multiple enlarged lymph nodes were also noticed at the root of the mesentery. On exploratory laparotomy, as no evidence of intussusception was found, no surgical reduction or bowel resection was performed, and the inflamed appendix was removed instead. No complication or recurrence of intussusception occurred after 3 years of the postoperative period.

Discussion

Acute appendicitis is the most frequent cause of abdominal surgical operations in emergency settings in pediatric and adult ages [3]. Intussusception is on the other hand primarily a childhood disease. Even if surgical investigation and treatment is essential in both of the conditions, intussusceptions may relieve without intervention in some cases [4]. Clinical signs and physical examination findings, in addition to almost typical presentation of periumbilical pain radiating to the right lower quadrant, mostly help to diagnose acute appendicitis. In some patients, however, the clinical findings may be obscure. Therefore, in such cases, imaging studies may be required to make a prompt diagnosis before perforation occurs. Today, this is achieved quickly and precisely by US and CT. US is the first-choice imaging technique in the preoperative diagnosis of acute appendicitis, particularly in pediatric patients, due to increasing concerns over radiation exposure with abdominal CT. Operator dependency may restrict the performance of ultrasound and bowel gas may obscure the vermiform appendix, especially when located behind the caecum and ascending colon. On the other hand, CT has been shown to be more sensitive and specific, when comparing the overall accuracy of US and CT [5]. CT should be the imaging modality of choice, particularly in overweight patients and when there is doubt of appendiceal perforation [3]. Choosing the appropriate imaging modality should be tailored uniquely for each patient.

Intussusception is defined as telescoping of a bowel segment and its mesentery into an adjacent bowel segment. In contrast to pediatric population, adult intussusception is less known, particularly due to its relative rarity and non-specific symptomatology accompanying variable clinical presentation. Thus, intussusception may require imaging in the event of inconclusive physical examination and laboratory findings.

Several imaging modalities have been proposed for the diagnosis of intussusception. While abdominal radiographs in erect position may be normal in early stages, air fluid levels due to small bowel obstruction may occur in advanced cases [6]. US is a preferential and reliable imaging modality in pediatric population and is also considered as

a useful first-step imaging tool in adults. US reveals a 'target' sign on transverse images and a 'pseudokidney' sign on longitudinal images. Recently, abdominal CT is regarded as the modality of choice in adult intussusception. 'Bowel within bowel' appearance on CT is virtually pathognomonic for intussusception [7]. CT is mandatory in adults not only to make diagnosis, but also to identify an underlying lead point and to investigate the complications.

By the virtue of its rarity, multiple intussusceptions are unexpected and US imaging may be terminated after recognizing an invaginated segment at a single site. Nevertheless, the intestinal tract should be carefully explored, keeping in mind the fact that there may be additional invaginated segments. The role of US may be restricted in this situation and CT is useful to explore the intestinal tract. Also CT may provide valuable findings of coexisting intraabdominal pathologies that can easily be overlooked by US, as it was the issue in the presented case.

Stopping the investigation after a satisfying finding to explain the cause of abdominal pain, may lead to some

diagnostic errors. In addition, irrelevant use of imaging techniques, especially CT, should be balanced by justification of the investigation, due to increasing concerns over potential stochastic effects of radiation exposure.

Adult intussusception is a rare phenomenon, and multiple transient intussusceptions are even more rare. Limited cases regarding the coexistence of intestinal malrotation with intussusception and inflammatory myofibroblastic tumor with acute appendicitis have been reported in previous studies [8,9]. This unique report adds precious clinical and imaging findings of acute appendicitis coexisting with multiple spontaneously resolving intussusceptions, to the literature.

Conclusions

Physicians should be alerted for accompanying multiple abdominal pathologies and use justification essentials to make their decisions about the selection of the appropriate imaging modality.

References:

1. Birnbaum BA, Wilson SR: Appendicitis at the millennium. *Radiology*, 2000; 215: 337–48
2. Aref H, Nawawi A, Altaf A et al: Transient small bowel intussusception in an adult: case report with intraoperative video and literature review. *BMC Surg*, 2015; 15: 36
3. Keyzer C, Gevenois PA (ed.), *Imaging of Acute Appendicitis in Adults and Children*, Medical Radiology, Diagnostic Imaging, Springer-Verlag Berlin; Heidelberg, 2011
4. de Clerck F, Pletinckx P, Claeys D et al: Intermittent adult ileocecal intussusception: a case report and review of literature. *Acta Clin Belg*, 2014; 69: 76–81
5. Hermanz-Schulman M: CT and US in the diagnosis of appendicitis: An argument for CT. *Radiology*, 2010; 255: 3–7
6. Byrne AT, Goeghegan T, Govender P et al: The imaging of intussusception. *Clin Radiol*, 2005; 60: 39–46
7. Kim YH, Blake MA, Harisinghani MG et al: Adult intestinal intussusception: CT appearances and identification of a causative lead point. *Radiographics*, 2006; 26: 733–44
8. Lukong CS, Maitama MI, Aminu B et al: Intestinal malrotation and midgut volvulus coexisting with intussusception. *S Afr J Surg*, 2007; 45: 30
9. Unver N, Coban G, Onaran OI et al: Co-existence of acute appendicitis and inflammatory myofibroblastic tumor of the small intestine: A case report. *Ann Med Surg*, 2015; 4: 217–20