PEDIATRIC ULTRASOUND



Intussusception: past, present and future

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Abstract Intussusception is a common etiology of acute abdominal pain in children. Over the last 70 years, there have been significant changes in how we diagnose and treat intussusception, with a more recent focus on the role of ultrasound. In this article we discuss historical and current approaches to intussusception, with an emphasis on ultrasound as a diagnostic and therapeutic modality.

Keywords Children · Fluoroscopy · Intussusception · Reduction · Technique · Ultrasound

Introduction

Intussusception is a common etiology of acute abdominal pain in children and is the most common cause of intestinal obstruction in young children [1]. Intussusception occurs when a segment of bowel (the intussusceptum) telescopes into an adjacent segment (the intussuscipiens). If untreated, intussusception results in ischemia, eventually leading to bowel necrosis, perforation and peritonitis.

Intussusception in children was nearly universally fatal until the nineteenth century [2]. The first successful air

Emily A. Edwards emily.edwards@ucsf.edu enema reduction was described in 1836 using "a common pair of bellows" [3], and the first successful surgical reduction in a pediatric patient was performed in 1871 [4]. By 1905, Harald Hirschsprung had accumulated 107 cases of hydrostatic enema reduction [5], but the treatment of intussusception in children was predominantly surgical until the mid-twentieth century. Articles published in the 1940s and 1950s demonstrated the efficacy and safety of enema reductions [6–8], which have remained the mainstay of treatment since.

Ultrasound (US) as a diagnostic modality for pediatric intussusception was first described in the early 1980s [9] and has since been widely adopted as the diagnostic test of choice for its high sensitivity and specificity, pathology characterization, and lack of ionizing radiation. Beyond diagnosis, the role of US in the treatment of intussusception remains a subject of active research in the pediatric literature and is discussed in detail later in this article.

Demographics

Intussusception is seen in children of all ages, with predominance in young children. More than 50% of intussusceptions occur in children younger than 1 year, with a nearly 2:1 male predilection [10–12]. Although older studies report seasonal variance in the incidence of intussusception, several newer studies have shown no seasonality [11–14], although some variation could be caused by geographic or demographic differences. Symptoms of colicky abdominal pain, bloody stool and a palpable abdominal mass are taught as the classic triad of intussusception but are present in less than 50% of children at the time of presentation [15]. Moreover there is significant symptomatic overlap with other acute abdominal conditions.

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Ileocolic intussusceptions represent the majority of cases (more than 80%), with the terminal ileum acting as the intussusceptum and the ascending colon as the intussuscipiens [16]. A "lead point" is thought to cause the bowel to invaginate and initiate the telescopic overlap in such a way that it gives rise to intussusception; however the majority of intussusceptions are idiopathic, with no clear lead point. Some of these idiopathic cases might be prompted by hyperplastic lymphoid tissue acting as the lead point in the distal small bowel (Fig. 1). In about 6% of cases, the intussusception lead point is a pathological lesion, such as a Meckel diverticulum, Henoch-Schönlein purpura, enteric duplication cyst, polyp or lymphoma [17, 18]. Iatrogenic intussusception has also been described with long enteral feeding tubes, with the tip of the feeding tube serving as a lead point (Fig. 2) [19].

Diagnosis

Diagnosing intussusception based on clinical presentation alone can be challenging. The symptoms of intussusception overlap with multiple other disease processes, and the inability of young children to communicate their symptoms compounds the clinical conundrum. Fortunately the imaging diagnosis of intussusception has been well described and can be made with a high degree of accuracy.

Enema

Historically, enema with barium, air or water-soluble contrast medium was used to diagnose a suspected intussusception. Contrast enema is a procedure familiar to all radiologists, one that used to be considered the standard diagnostic test for intussusception. Introducing contrast material into the colon shows the intussusception as an intraluminal crescent or round filling defect (Fig. 3) [8], and the location and imaging appearance of the leading edge of the intussusception can predict the likelihood of successful reduction [20]. Relative shortcomings of diagnostic enema include limited detection of other causes of acute abdominal pain, low sensitivity for small-bowel intussusceptions, invasive technique, and exposure to ionizing radiation. One advantage of diagnosing intussusception by enema is the possibility for immediate conversion to a therapeutic procedure.

Ultrasound

Over the last two decades, US imaging has emerged as the first-line modality for the diagnosis of a suspected intussusception. It has high sensitivity and specificity for detection of intussusception (97.9% and 97.8%,



Fig. 1 Idiopathic ileocolic intussusception. Longitudinal abdominal US in a 3-year-old girl with idiopathic ileocolic intussusception containing lymph nodes (*arrows*). Lymph nodes are commonly present and might prompt intussusception by acting as a lead point for invagination of bowel

respectively), as well as a high negative predictive value (99.7%) [21]. Given that 86% of pediatric patients with suspected intussusception have negative sonograms [21], these children can be spared an unnecessary and invasive enema when US is used as the initial imaging test, and conditions mimicking intussusception can be identified and more appropriately triaged. Furthermore, the lack of ionizing radiation makes US well suited to image pediatric patients.

The US appearance of a typical ileocolic intussusception is highly characteristic and has been well described in the literature. A peripheral hypoechoic ring with central echogenicity, known as the target sign (in transverse view) and pseudokidney sign (in longitudinal view), correspond to the bowel wall surrounding hyperechoic mesenteric fat contained within the intussusception (Fig. 4) [22]. Most commonly an ileocolic intussusception is

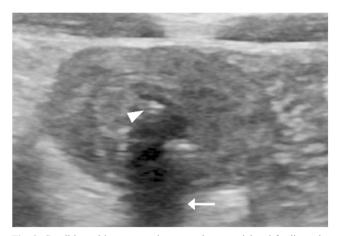


Fig. 2 Small-bowel intussusception around a gastrojejunal feeding tube in a 2-year-old girl. Transverse abdominal US shows an echogenic center (*arrowhead*) with posterior shadowing (*arrow*), which indicates the presence of the feeding tube within the intussusception

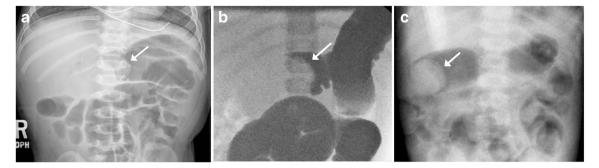


Fig. 3 Ileocolic intussusception. The leading edge of the intussusception appears as an intraluminal round or crescent filling defect (*arrows*), a classic finding of ileocolic intussusception on multiple modalities. **a**

Anteroposterior radiograph in a 7-month-old boy. **b** Water-soluble contrast enema in an 11-month-old boy. **c** Air contrast enema in a 10-month-old girl

found in the right abdomen with a diameter of 2–4 cm. The large size both facilitates detection and helps distinguish ileocolic intussusception from a more diminutive small bowel–small bowel intussusception [23].

Doppler US contributes to the diagnosis of intussusception in several ways. Absence of blood flow within the intussusception has been shown to correlate with bowel ischemia and necrosis at surgery and is a predictor of unsuccessful enema reduction (Fig. 5) [24]. It is an important finding in conjunction with the overall clinical status of the child in determining appropriate management. Doppler blood flow characteristics are also useful for differentiating between possible pathological lead point lesions.

In addition to Doppler features, multiple other US findings can be used to predict the likelihood of success at subsequent enema reduction. These findings — which might reflect an incarcerated intussusception — include free intraperitoneal fluid, pneumatosis, fluid trapped within the intussusception (Fig. 5) and small-bowel obstruction. One series reported an overall successful reduction rate of 72%, which increased to 93% in the absence of these negative predictive signs and was as low as 25%when trapped fluid was seen between the colon and the intussusceptum [25]. When seen, these negative predictive signs are important for counseling children and their parents and can influence how the radiologist approaches the enema reduction with the expectation that it might be unsuccessful and have a higher chance of perforation.

Abdominal radiographs

The role of abdominal radiographs in the diagnosis of intussusception is somewhat controversial. Radiographs are insensitive for detection of intussusception, with one series indicating that only 45% of intussusceptions were correctly identified on abdominal radiographs [26]. Some authors have advocated use of abdominal radiographs as a problem-solving tool only, such as suspicion for bowel perforation with signs and symptoms of peritonitis, atypical clinical presentation, or an equivocal US examination [15, 27]. Although abdominal radiographs do add diagnostic value in many cases, the poor test characteristics compared to US imaging suggest that they should not be used as a primary screening test for suspected intussusception, although they might be performed prior to reduction to exclude pneumoperitoneum [28].

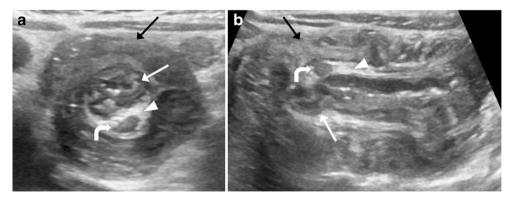


Fig. 4 Classic US appearance of an ileocolic intussusception. US images of the right upper quadrant in a 10-month-old girl. **a** Transverse image shows the target sign, with the hypoechoic colon (*black arrow*)

containing the intussusceptum (*white arrow*), and mesenteric fat (*arrowhead*) and lymph nodes (*curved arrow*). **b** Longitudinal image shows the pseudokidney sign with the same structures

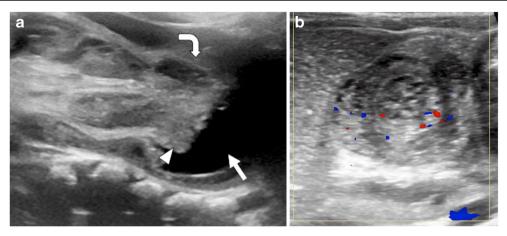
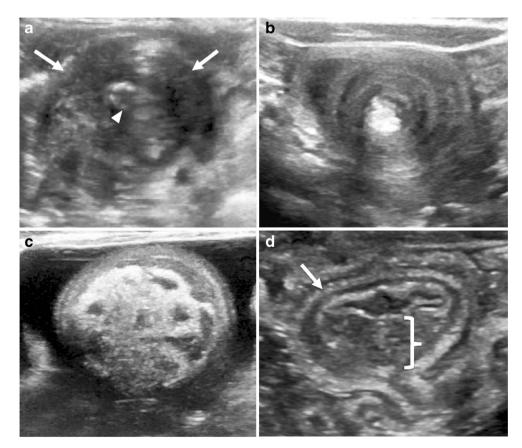


Fig. 5 US imaging findings in predicting likelihood of enema reduction and perforation risk. **a** Longitudinal image in a 3-month-old boy shows trapped fluid (*arrow*) within the intussuscipiens (*curved arrow*). The leading edge of the intussusceptum (*arrowhead*) extends nearly to the

rectum and was not reducible by enema. **b** Transverse image of the right upper quadrant in a 2-year-old boy with 2 days of intermittent abdominal pain shows minimal bowel wall Doppler blood flow, which correlates with bowel ischemia and predicts less successful reduction

Mimics

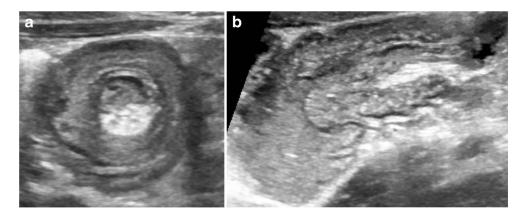
Because the clinical presentation of intussusception can be nonspecific, the radiologist must consider a broad differential diagnosis for the child's acute abdominal pain. An advantage of using US imaging is that it allows for evaluation and diagnosis of a variety of causes of acute abdominal pain in children, including intussusception. As with any diagnostic test, there are important pitfalls and mimics to be aware of when interpreting a US examination for suspected intussusception. The false-negative rate for US is low, and non-visualization of an intussusception on US imaging has a negative predictive value of 100% [21, 29]. Several entities can mimic intussusception on US, including appendicitis, Meckel diverticulum, small bowel–small bowel intussusception and stool (Figs. 6 and 7). Perforated appendicitis is a particularly important



intussusception on abdominal US imaging. a Transverse image of the right upper quadrant in a 2year-old boy with pancreatitis shows a rounded peripancreatic collection (arrows) with central echogenic tissue (arrowhead). b Transverse image in a 10-monthold boy with a small-bowel intussusception from an inverted Meckel diverticulum shows concentric rings of hypoechoic bowel and central echogenicity. c Transverse image in a 5-week-old boy with inspissated stool shows echogenic bowel contents with scattered ovoid hypoechoic foci, which could be mistaken for mesenteric fat and lymph nodes. Ascites is also present. d Transverse image in a 4-year-old boy with terminal ileitis shows an edematous ileocecal valve (bracket) within the thickened cecum (arrow)

Fig. 6 Mimics of ileocolic

Fig. 7 Small bowel–small bowel intussusception in a 2-year-old boy. Transverse (a) and longitudinal (b) US images demonstrate a small bowel–small bowel intussusception. Size is an important distinguishing feature; this intussusception measured less than 2 cm in transverse diameter, smaller than a typical ileocolic intussusception



mimic of intussusception because the perforated appendix and adjacent contained collection can have the classic appearance of alternating concentric layers of hyper- and hypoechoic tissues with an echogenic center, with enteric contents or an appendicolith mimicking the mesenteric fat within an intussusception (Fig. 8) [30, 31].

A highly specific US finding of intussusception is the presence of mesenteric lymph nodes within the lumen of the intussuscipiens (Fig. 1) [23, 30]. Identifying lymph nodes within the lesion in question strongly suggests intussusception as the diagnosis, and might help distinguish between true intussusception and its mimics. This is especially true in the distinction between ileocolic and small bowel-small bowel intussusceptions, which do not contain lymph nodes or mesenteric fat and are typically less than 2 cm in diameter. Additionally, in our experience idiopathic small bowel-small bowel intussusceptions are often transient. If one is detected during a US examination, rescanning the area of interest at the end of the examination usually demonstrates resolution of the small bowel-small bowel intussusception, while a symptomatic ileocolic intussusception typically persists. Attention to these details can greatly improve diagnostic confidence.

Treatment

Since its wide implementation in the mid-twentieth century, enema reduction remains the gold standard for nonsurgical treatment of intussusception. Over the last several decades there have been changing trends in both enema material and imaging modality for intra-procedural monitoring [32–34], but the core concept — introducing pressurized air or liquid into the colon to push the intussusceptum back through the ileocecal valve with real-time imaging observation — is the same for all techniques.

Enema material

Barium was the contrast agent of choice for intussusception reduction when the procedure was initially introduced, as a therapeutic conversion of the diagnostic barium enema. Barium has subsequently become less popular because of concerns over peritoneal staining in the event of perforation [35, 36]. Currently pneumatic enema or hydrostatic enema with saline or water-soluble contrast are the dominant techniques, with a recent survey of North American pediatric radiologists indicating that 78% of respondents use air to reduce

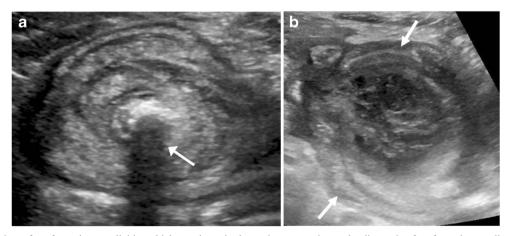


Fig. 8 US imaging of perforated appendicitis, which can have both clinical and imaging overlap with intussusception. a Right lower quadrant transverse US image in a 10-year-old girl shows central shadowing (*arrow*), indicating the presence of an appendicolith, an

important clue to the diagnosis of perforated appendicitis. **b** Right lower quadrant transverse image in a 4-year-old girl shows a rounded periappendiceal collection with surrounding alternating hyper- and hypoechoic tissue (*arrows*) that can mimic intussusception

intussusceptions [34], compared to 85% using barium in 1989 [37] (air reduction was first described in the late 1980s [38, 39]). Numerous studies have compared air and liquid methods, with a recent meta-analysis of more than 32,000 children indicating superior success and similar perforation rates with air enema reduction when compared to hydrostatic reduction [40]. In addition to higher success rates, air enema is also cleaner, less expensive, and has the added advantage of shorter fluoroscopic times and lower radiation exposure [33].

Imaging modality

The best imaging modality to use when monitoring an intussusception reduction is a subject of active investigation and debate in the pediatric literature. Historically, fluoroscopy has been the preferred imaging modality for intussusception reduction, favored for its familiarity and a large body of supporting evidence. While fluoroscopic monitoring remains a widely used technique, the conversation around nonoperative management of intussusception has increasingly involved the role of ultrasound. Studies dating to the 1980s have shown similar success rates for sonographic reduction when compared to fluoroscopic reduction [41-44], including a recent head-tohead comparison of the two techniques [45]. Despite the growing evidence in support of sonographic reduction, a 2015 survey found that only 4% of North American pediatric radiologists used US to monitor intussusception reductions [34]. Advocates for sonographic reduction cite the lack of ionizing radiation and characterization of possible pathological lead points as distinct advantages of using US. However availability of sonographers after hours and comfort with sonographic reduction are some barriers to more widespread implementation.

Other considerations

An additional technique that has been described for the nonsurgical management of intussusception is external manual compression as a means of reduction. Using directional, graded compression and intermittent observation with US imaging, Vazquez et al. [46] reported a primary reduction rate of 80% with no perforations — without ionizing radiation — and the noninvasive nature of the intervention results in rapid post-procedure recovery [46]. While certainly less well studied than other reduction methods, external manual compression warrants further investigation as a noninvasive approach to the conservative treatment of intussusception in appropriately selected patients.

Repeated, delayed attempts at reduction have also been described in the literature, with improved success rates of 50– 82% [27]. The different reduction techniques studied, and particularly the variability in time elapsed between repeat attempts (from 30 min to more than 24 h), limits extrapolation of the data in some scenarios; however in children who remain clinically stable and in whom the intussusceptum was at least partially reduced on the prior attempt it might be worthwhile to repeat reduction with the goal of avoiding unnecessary surgery [47].

Complications

The feared complication of intussusception is bowel perforation. Currently the mean perforation rate for intussusception reduction is reported at 0.8% [27], with similar perforation rates for pneumatic and hydrostatic reduction. On average, a pediatric radiologist has a 34% chance of encountering a perforation once in a 10-year period [34]. If a perforation occurs during air reduction, there is potential for tension pneumoperitoneum and cardiovascular collapse, a rare but life-threatening complication. Overall, because of the safety and efficacy of treatments for intussusception the mortality rate is extremely low — 2.1 per 1 million live births [48].

Institutional practices

At our institution, we use a combination of US imaging and fluoroscopy in the diagnosis and treatment of routine intussusception. We perform a diagnostic sonography or We perform a diagnostic sonogram for all children with suspected intussusception, including transfers from outside institutions, because of the relative frequency of spontaneous reduction (13%) [13]. If an ileocolic intussusception is confirmed by US, the pediatric surgery service is notified, and the radiologist obtains informed consent from the child's parents. Discussion with the parents includes risks such as perforation, the perceived likelihood of successful enema reduction based on the clinical presentation and sonographic appearance, and the need for surgery if enema is unsuccessful. We establish intravenous access prior to starting the procedure, and we include an angiocatheter needle on the procedure tray for decompression in the event of tension pneumoperitoneum. We do not use antibiotics, analgesia or sedation. Our primary enema reduction technique is fluoroscopically guided air reduction, with water-soluble liquid contrast material on an as-needed basis, and a maximum insufflation pressure of 120 mmHg with a pop-off valve for added safety. Children with successful intussusception reductions are admitted for observation and to monitor for recurrence, while those with unsuccessful reductions are referred to pediatric surgery for further management.

Discussion

There have been major changes over the last century to how pediatric intussusception is diagnosed and treated. While these changes have had a significant positive impact on intussusception mortality rates, there is still work to be done as we seek to further refine our methods and improve patient outcomes. With respect to diagnostic modalities, the literature over the last several decades reveals broad support for US imaging as the diagnostic test of choice for suspected intussusception. Ultrasound has proved to be an effective imaging test for diagnosis, with high sensitivity and specificity, high negative predictive value, no ionizing radiation, and utility in characterizing pathological lead point lesions, predicting likelihood of successful reduction, and evaluating for other possible causes of abdominal pain. As a result, practices have moved away from diagnostic enema and abdominal radiography except in select cases.

Several techniques are used for intussusception reduction. In comparing current practice patterns to surveys conducted in the 1990s, a few general trends in treatment of intussusception have emerged [32, 34, 49, 50]. A majority of enema reductions are now performed with air, whereas 25 years ago a majority were performed with barium. Practitioners have gravitated toward pneumatic reduction for its equivalent efficacy to hydrostatic reduction while being cleaner and cheaper and having lower radiation doses when compared to contrast enema.

Despite the changes in favored diagnostic modality and enema reduction contrast material over the last three decades, there is a continued strong preference among North American pediatric radiologists for fluoroscopy over sonography to monitor intussusception reduction. Numerous studies have shown sonographic monitoring of intussusception reduction to be as effective as fluoroscopic monitoring but without exposing the child to ionizing radiation. However a recent survey indicates that fewer than 5% of intussusception reductions are monitored with US, suggesting that although the literature support its use there are barriers to wider implementation.

Thus far the overwhelming majority of studies published on sonographic reduction of intussusception have used a liquid enema material. Meanwhile, over the same period of time, pediatric radiologists have increasingly favored air as the preferred medium for enema reduction, for all of the reasons discussed. This trend toward air reductions might discourage some practitioners from adopting US monitoring in order to avoid the relative disadvantages of liquid enemas. With the aim of evaluating whether the benefits of pneumatic reduction could be combined with radiation-free sonographic monitoring, a 2001 series demonstrated feasibility of the technique with a high success rate (92%) but also a relatively high perforation rate (4% compared to 0.14-2.80%), which was attributed to a technical learning curve [51]. Whether pneumatic reduction and US monitoring can be used in conjunction safely and effectively in a larger, randomized study population is an important area for further investigation.

A fair criticism of the literature on sonographic monitoring of intussusception reduction, or of any nonsurgical reduction technique, is the lack of multicenter, prospective, randomized trials to determine the relative efficacy and safety of the various techniques in use. Small prospective randomized studies have been performed [52] but not at a scale sufficient to standardize practices, particularly at the international level where there is significant discrepancy in reduction methods.

Finally, an essential consideration is the comfort of individual practitioners with a particular technique, a phenomenon that is hardly unique to intussusception reduction. Given the small number of North American pediatric radiologists who use sonography to monitor reductions, this implies a similarly small number of residents and fellows who are exposed to the technique during training. Without a critical mass of radiologists who are comfortable with sonographic monitoring, changes in practice patterns will be slow.

Conclusion

Expanding use of US imaging in the diagnosis and treatment of pediatric intussusception has numerous benefits. In many parts of the world, fluoroscopy is impractical because of limited access, and increasing provider familiarity with sonography could provide safe, effective options for diagnosing and treating intussusception in these regions. Furthermore, the ALARA (dose as low as reasonably achievable) principle mandates that we strive to reduce or eliminate medical radiation when safe and feasible. Management of intussusception, one of the most common abdominal emergencies in children, provides an opportunity to implement this principle at the individual, institutional and international levels. Medical advancements have changed intussusception from a disease that was most often fatal to one that is easily diagnosed and effectively treated. By building on the efforts of the last 100 years, we can continue to make the diagnosis and treatment of intussusception safer and less morbid for pediatric patients everywhere.

Compliance with ethical standards

Conflicts of interest Dr. J. D. MacKenzie receives research grant support from GE Healthcare. Drs. Edwards, Pigg, Courtier, Zapala and Phelps declare no financial interests. The authors have no investigational or off-label uses to disclose.

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