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Case Report

Unusual ultrasound appearance of small bowel intussusception and secondary bowel obstruction in a child with Peutz-Jeghers syndrome

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ABSTRACT

Small bowel intussusception (SBI) in pediatric patients resolves spontaneously in the majority of cases. Pathologic small bowel intussusception with a lead point is rare in children. Ultrasound (US) is the preferred initial imaging study for the diagnosis of intussusception. We report a case of long-segment SBI and secondary bowel obstruction caused by a large hamartomatous polyp.

This case emphasizes unique, atypical ultrasound findings that may be encountered in small bowel intussusception, with correlative radiographic, CT (computed tomography) and intra-operative findings. Increased awareness of these atypical imaging features can lead to early diagnosis and decrease the risk of potential complications including mesenteric venous thrombosis, bowel ischemia and necrosis.

1. Introduction

Small bowel to small bowel intussusception (SBI) is uncommon compared to ileocolic intussusception, and its incidence ranges from 0.4% to 17% of all intussusceptions in children [1,2]. The rate of spontaneous reduction is high, ranging from 57% [3] to 100% [1,2], and a pathologic lead point is found in up to 69% of the patients with persistent symptoms who undergo surgical exploration [4]. Typical sonographic and CT appearance of a spontaneously reducing SBI is a small diameter short-segment intussusception frequently located in the left abdomen or paraumbilical region [1,5]. We present a case of massive jejuno-jejunal intussusception caused by a large infarcted Peutz-Jeghers polyp, with atypical clinical presentation and ultrasound (US) features.

2. Case

A 5-year-old male presented to the emergency department (ED) with acute on chronic recurrent vomiting, acutely worsening abdominal pain, and severe dehydration for one day. The patient had intermittent, spontaneously resolving episodes of postprandial vomiting and abdominal pain for approximately 4 months. On presentation, laboratory data showed metabolic acidosis, leukocytosis, acute renal insufficiency, as well as chronic anemia. Abdominal US (Fig. 1) demonstrated multiple non-dilated bowel loops surrounded by complex fluid with echogenic debris and septations, contained in a thick-walled structure extending from the left upper quadrant to the right lower quadrant. Abdominal radiograph (Fig. 2) showed paucity of bowel gas in the mid abdomen with a large central dense structure causing mass effect on the adjacent ascending and transverse colon, and displacing them laterally and superiorly, respectively. CT examination of the abdomen and pelvis with oral and intravenous contrast was obtained 12 h after admission following recovery of the renal function with hydration. This exam revealed a high grade small bowel obstruction with a massively distended proximal jejunal loop containing both dilated and non-dilated small bowel loops, mesentery, and extraluminal fluid corresponding to the findings seen on the admission US (Fig. 3). The dilated jejunum (intussuscipiens) showed substantial wall thickening with edema and mural hypoenhancement, concerning for ischemia. Patient was immediately taken to the operating room (OR) for exploratory laparotomy. Intraoperatively, the patient was noted to have a nonreducible jejuno-jejunal intussusception causing obstruction and bowel ischemia (Fig. 4A), with approximately 40 cm of devitalized bowel. A 4 cm infarcted polyp was serving as the lead point for the intussusception (Fig. 4B).

A segmental bowel resection of the infarcted segment inclusive of the polypoid lead point with a primary small bowel end-to-end anastomosis was completed. The ischemic necrosis throughout the polyp limited histologic assessment. However, the dominant histologic features in the available viable tissue were most consistent with a Peutz-Jeghers

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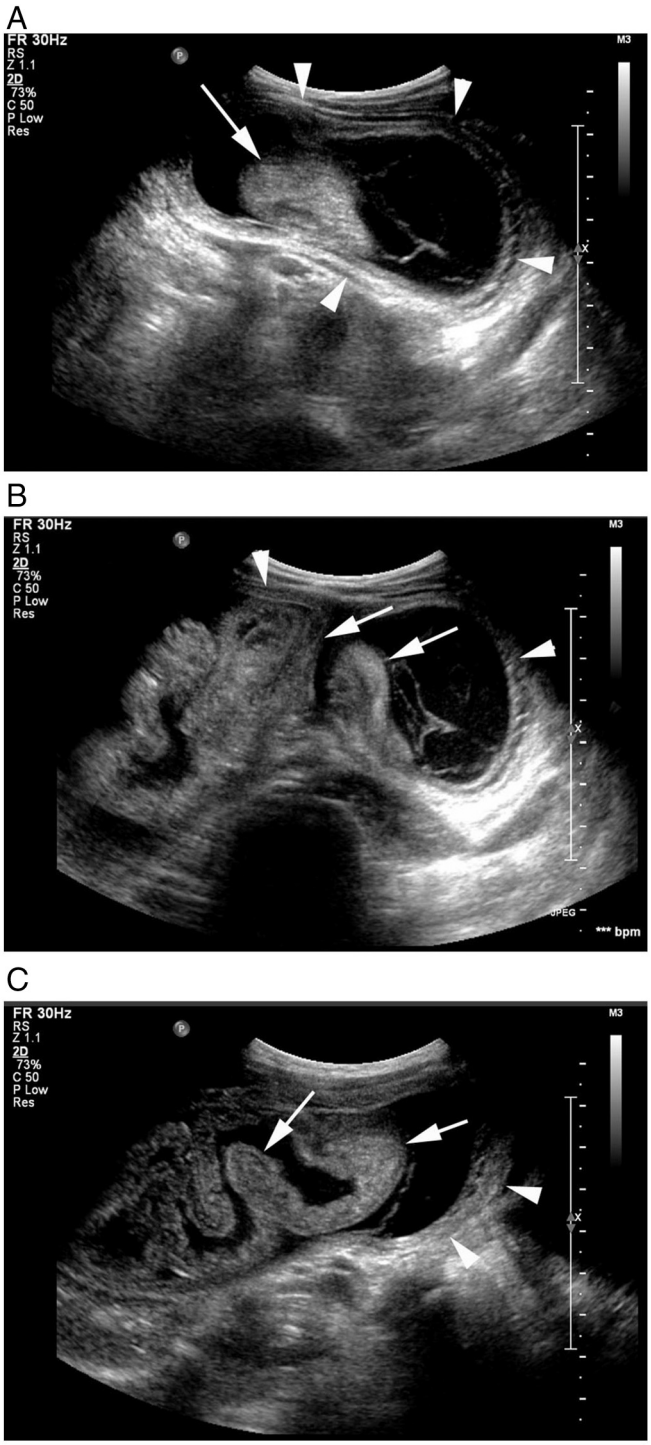

Fig. 1. Transverse (A and B) and longitudinal (C) grayscale ultrasound images of the left lower quadrant demonstrating multiple decompressed small bowel loops (arrows) with anechoic adjacent fluid containing echogenic septations, encased in a thick-walled structure (arrowheads) extending across the abdominal cavity.

polyp. No additional polyps were found in the resected bowel. The child had an uneventful postoperative recovery.

3. Discussion

Intussusception is the second most common cause of acute abdomen in children, with ileocolic intussusception representing up to 90% of the cases. In the pediatric population, SBI is uncommon and accounts for 0.4% to 14.2% of all intussusceptions [1,2]. Patients who present with

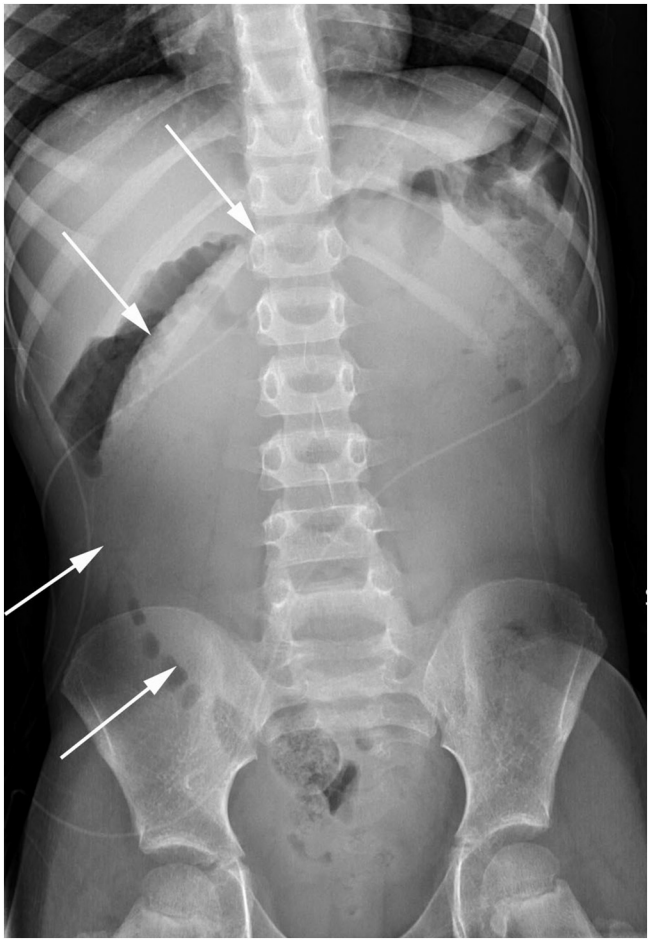

Fig. 2. Supine abdominal radiograph demonstrates an overall paucity of bowel gas and a large dense structure in the central abdomen (arrows) exerting mass effect and displacing the ascending and transverse colon segments laterally and superiorly, respectively.

SBI are generally older, with a mean age of 4.6 years in one series [6] and 11.2 years in another [1], while ileocolic intussusceptions tend to affect patients that are younger than those with symptomatic small bowel intussusception, commonly less than 2 years of age [6–8]. In contradistinction to ileocolic intussusception, there is also a high rate of spontaneous SBI reduction ranging from 57% to 100% [1–3]. Clinical presentation is often non-specific and may include vomiting, abdominal pain, fever, bloody/currant jelly stool, and infrequently a palpable abdominal mass [5].

Typical US findings of SBI include: a doughnut-like lesion (hypoechoic outer rim surrounding a round hyperechoic center), hyperechoic crescent surrounded by a hypoechoic rim, and multiple concentric rings of alternating hypo- and hyper-echogenicity [5]. While ileo-ileal intussusception can be found near the ileocecal valve or in the right lower quadrant, both jejuno-jenunal and ileo-ileal intussusceptions are more commonly located in the left abdomen or paraumbilical region. Small bowel intussusceptions usually measure 2–3 cm in diameter [5]. CT findings are similar to sonography with a small caliber layered- or targetoid-appearing mass which may contain mesenteric fat [1]. By contrast, ileocolic intussusceptions are generally larger in diameter, measuring 3–5 cm, and typically located in the right lower or right upper abdominal quadrants. Imaging characteristics which favor spontaneous reduction of SBI include: 1. length less than 3.5 cm; 2. visible peristalsis; 3. thin outer wall (3.3 mm in spontaneously reduced SBI vs 6.8 mm in SBIs requiring surgical reduction); and 4. absence of a lead point [4,9]. Common lead points encountered during surgical reduction include hamartomatous polyps in Peutz-Jeghers syndrome, Meckel's diverticulum, enteric duplication cyst,