Acute postappendicectomy hemorrhage due to isolated cecal necrosis in a 9-year-old boy: A case report

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ABSTRACT
Primary inflammatory processes of the cecum (typhlitis) were originally described in children with neutropenia or immunosuppression. It has also been described in the elderly population as an infrequent variant of ischemic colitis. We present a case of a previously healthy 9-year-old boy who presented with right lower quadrant abdominal pain, low grade fever and localized right iliac fossa tenderness. At open appendicectomy his appendix was noted to be mildly congested only. On the 2nd postoperative day the patient had a significant per rectal bleed, and exhibited signs of peritonitis. He underwent an emergency laparotomy. The appendicular stump tie was found intact with no active bleeding from the stump. There were large clots and fresh bleeding identified within a necrotic cecum. A cecrectomy was performed with an end-to-end anastomosis. He made an uneventful recovery and remains well 6 months post-operatively. The histopathologic examination of the specimen’s confirmed focal mucosal inflammation of the appendix and extensive transmural necrosis of the cecum. Cecal necrosis must be considered in any patient presenting with signs of appendicitis with per rectal bleeding.

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1. Case report

A 9-year-old boy presented to the emergency department with a 48 h history of right lower quadrant abdominal pain and fever without associated vomiting or diarrhea. He had no significant background medical history. On admission he was tachycardic (112/min), pyrexial (37.7 °C) and had localized RIF tenderness with no guarding. His laboratory data revealed a leukocyte count of 5.4/μL (normal range 4.0–10.8/μL) and a C-reactive protein of 20.8 mg/L (normal range 0–10 mg/L). He was admitted for observation. He deteriorated clinically overnight and the following morning had an abdominal ultrasound which was within normal limits. A clinical diagnosis of acute appendicitis was made, the child was commenced on amoxicillin/clavulanic acid and was brought for surgery.

At surgery the appendix was mildly congested but there was no other cause found for the child’s symptoms. An appendicectomy was performed. On the 2nd postoperative day the child started to vomit and passed fresh blood per rectum with diarrhea. His abdomen became distended with generalized tenderness and guarding. A nasogastric tube was passed, fluid resuscitation commenced, repeat blood tests including coagulation screen were performed and metronidazole and gentamicin was added to his antimicrobial regime. The child became tachycardic despite adequate fluid...
resuscitation and had a 3 g drop in his hemoglobin. He was returned to theater with a suspected diagnosis of appendix stump bleeding.

At laparotomy, the appendicular stump tie was found intact with no active bleeding from the stump. Cecum externally appeared congested, but otherwise normal with no evidence of intraperitoneal bleeding. The appendix stump and base of cecum were opened. Large clots and fresh bleeding were identified within the cecal lumen. No specific bleeding point was noted, and the entire mucosal surface appeared necrotic and hemorrhagic. The cecum including the ileocecal valve was removed, and an ileo-colic end-to-end anastomosis was performed. Postoperatively he was transferred to high dependency unit. He was commenced on diet on postoperative day three and was discharged home on postoperative day six. The histopathologic examination of the appendix confirmed focal mucosal inflammation only; the cecal specimen revealed transmural necrosis with extensive mucosal ulceration, transmural suppuration and hemorrhage. These appearances were reminiscent of necrotizing enterocolitis and there was no evidence of primary mesenteric vascular pathology (Fig. 1). The child remained well and was discharged from the out-patients clinic after 6 months.

2. Discussion

Typhlitis, also known as neutropenic enterocolitis or ileocecal syndrome, refers to the localized inflammation of cecum primarily found in neutropenic patients with a high associated mortality [1,3,4]. This condition is typically seen in children and adults with hematological malignancies who are neutropenic and underwent induction cytotoxic chemotherapy causing breakdown of gut mucosal integrity. Typhlitis should be considered in any neutropenic or immunosuppressed individual presenting with fever, abdominal pain and RIF tenderness [5]. As our child was previously healthy with an acute onset of right lower quadrant abdominal pain, typhlitis is an unlikely explanation.

Acute primary cecal necrosis has been already reported in immunocompetent adults as an infrequent variant of ischemic colitis [2], as well as a rare form of adult necrotizing enterocolitis (ANEC) and nonocclusive mesenteric ischemia [6]. Acute colonic ischemia is the most common cause of colitis in the elderly population [7]. Focal ischemia or infarction isolated to the cecum is a rare entity; with only a few cases reported to occur in adults spontaneously [2], and in association with chronic heart disease [8], open-heart surgery [9], hemodialysis [10], and systemic hypotension due to or trauma [11]. Such an explanation is unlikely in our child, because of the lack of predisposing factors; and there was no vascular thrombosis or abnormality noted during the pathological analysis of the resected segment of bowel.

As the bleeding occurred 30 h following appendicectomy, the possibility of surgical intervention itself as the cause of this bleeding must be considered. Most early postoperative significant bleeding following appendicectomy are related to stump suture becoming loose or undone with resultant stump bleeding [12,13]. However, at the time of surgery, the stump suture was noted to be intact with no oozing from the stump or blood clots within the immediate surroundings of the stump in the peritoneal cavity. The cecal mucosal ulceration and bleeding were only noted after removal of the stump suture and exploration of the intraluminal surface of cecum by opening the appendix stump and cecal wall further. We considered the possibility of excess diathermy use along the serosa of the cecum causing necrosis and bleeding from the cecal wall. There was no serosal burning or damage noted at the time of surgery, or during the histological analysis of resected cecum.

Diagnosis in acute cecal necrosis can be very challenging, because patients with this condition usually present with right lower quadrant abdominal pain associated with elevated laboratory inflammatory markers suggestive of acute appendicitis [14]. Abdominal ultrasound and Computed Tomography has been reported helpful; cecal wall thickening with isolated pneumatisis coli are nonspecific findings, however strongly suggestive of the diagnosis [15]. Treatment of isolated cecal necrosis is surgical; right hemicolectomy, cecal resection with ileocolostomy as well as laparoscopic partial cecal resection for partial cecal necrosis have been reported with satisfactory results [16].

3. Conclusion

Isolated cecal necrosis is a rare entity in adults and to our knowledge has not been reported in a previously healthy child yet. When significant rectal bleeding together with signs of appendicitis is observed, this rare condition must be included within the differential diagnosis.

Consent

Written informed consent was obtained from the patient’s father (as legal guardian) for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest

The authors have none to declare.

References


Fig. 1. Low power photomicrograph showing full thickness hemorrhagic necrosis of the cecal wall.