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Congenital cecal diverticulitis in a pediatric patient

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Abstract

Diverticulitis in the pediatric population is a very rare cause of abdominal pain. When present in the cecum or ascending colon, it is often incorrectly diagnosed preoperatively as acute appendicitis. This is especially true in Western countries where right-sided diverticulitis is less common. Here we detail a case of a pediatric patient with complicated congenital cecal diverticulitis and review the literature on pertinent management. An extensive work up with imaging and endoscopy was completed and definitive surgical treatment with diverticulectomy an appendectomy was performed. As the incidence of diverticular disease in younger individuals increases, right sided diverticulitis is worthy of consideration on the differential diagnosis.

Keywords

Cecal diverticulitis; Congenital diverticulum; Pediatric

1. Case report

JL is an 11-year-old Caucasian female with a past medical history of Irritable Bowel Syndrome who complained of 3 days of sharp, worsening right lower quadrant abdominal pain. The patient denied fever, nausea, vomiting, hematochezia, and diarrhea but endorsed increased frequency of straining with bowel movements which were resolved with polyethylene glycol. She was transferred to a pediatric hospital following computed tomography (CT) scan of the abdomen and pelvis that showed a large colonic diverticulum

Patient consent

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All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

with findings suggestive of acute diverticulitis. Her white blood cell count (WBC) prior to transfer was 11.7 k/cumm, while WBC on admission was 8.4 k/cumm. On physical examination, the abdomen was soft and nondistended and bowel sounds were present. There was involuntary guarding and rebound with severe tenderness to palpation to the right lower quadrant. Psoas, Obturator, and Rovsing signs were negative. The patient was admitted on a clear liquid diet with adequate pain control and started on maintenance IV fluids and antibiotics including piperacillin-tazobactam.

On day 1 of admission, her CT scan was reviewed with dedicated pediatric radiologists. Findings were felt to most likely represent "Crohn disease versus nonspecific focal colitis/ enteritis," without diverticulosis or diverticulitis (Fig. 1). Esophagogastroduodenoscopy and colonoscopy were planned on day 2 which grossly showed normal appearing esophagus, stomach, duodenum, terminal ileum, and entire colon (Fig. 2). Biopsies were taken of the terminal ileum and cecum which showed normal mucosa without acute inflammation or granulomas. Fecal calprotectin at this time was normal (57 mGg/gm) and antibiotics were discontinued due to less concern for infection. Pelvic ultrasound (US) was performed in order to rule out ovarian pathology and this was normal. On the 3rd and final day of admission, the patient had a magnetic resonance enterogram (MRE) of the small bowel after varying reads of the CT scan and inconclusive endoscopy studies. MRE was most suggestive of anteromedial congenital cecal diverticulitis (Fig. 3). The patient was discharged in the evening and started on a 14-day course of ciprofloxacin and metronidazole for treatment of acute diverticulitis.

On outpatient follow up with Pediatric Surgery, the patient had continued but much improved right-sided abdominal pain and was eating, drinking, and stooling without difficulty. The plan at this time was to wait 6 weeks after discharge to allow the inflammation to subside and then perform laparoscopic appendectomy and cecal diverticulectomy for definitive treatment. This decision was made as there was originally phlegmon present despite imaging that suggested a normal appendix and the risk of recurrence was determined to be high. The patient was admitted for bowel prep the day prior to the scheduled surgery. The patient was taken to the operating room where a transumbilical and 2 lower abdominal incisions were made for port placement per standard fashion for laparoscopic appendectomy. The diverticulum at this point was not immediately identified and the right colon was mobilized. After taking down the white line of Toldt, mobilizing the colon medially, and still not identifying the diverticulum the periumbilical incision was enlarged and the cecum was eviscerated and inspected in an open fashion. The fat was taken down in an area of visible inflammation immediately adjacent to the appendix on the anteromedial cecum which revealed a small 1 cm diverticular appearing structure consistent with MRE findings. The diverticulum was amputated (Fig. 4) and closed in two layers with a running 3-0 Vicryl, followed by multiple interrupted 3-0 silk sutures. It was decided that removing the appendix was also warranted. The appendiceal mesentery was taken down with cautery, followed by placement of 2 separate 3-0 Vicryl ligatures at the base of the appendix prior to amputation. No other findings were identified on the cecum, which was returned to the abdominal cavity and the incisions were appropriately closed. Final pathologic description of the specimen confirmed cecal diverticulum and normal appendix

(Fig. 5). The patient was discharged the following day with well controlled pain, tolerating oral intake, and ambulating without difficulty.

2. Discussion

Diverticula are commonly thought to be an acquired malformation that arise from a chronic increase in intraluminal pressure. This is supported by a number of modifiable risk factors that increase the incidence of the disease with an increase in age including obesity, low-fiber diet, and physical inactivity. This makes a diverticulum of congenital etiology, which may present with complications relatively younger in life, very rare. Presenting with inflammation or infection of the diverticulum is even more rare, considering that less than 5% of all patients with diverticula of any etiology develop diverticulitis [1]. Although approximately 65% of the Western population has diverticulosis by 85 years old, the prevalence in the population younger than 40 is only 10% [2]. The highest increase in incidence of diverticulitis within recent decades falls within this age group. Even so, the diagnosis of diverticula in the pediatric patient may only happen during work up for abdominal pain or operative treatment of the acute abdomen and the true incidence may not be known. No matter the age group or etiology, diverticula can develop anywhere along the colon from the cecum and appendix to the sigmoid colon. In Western countries, it is much more common to have diverticula of the left colon as compared to the cecum and ascending colon [3]. In North America it is estimated that only 1–2% of all cases are on the right, however, there appears to be geographical differences in distribution of diverticular disease. Right sided disease may be just as prevalent in Asian countries with 43-50% of all cases arising from this more proximal portion [3]. Ultimately these factors make cecal diverticulitis in the pediatric population a rare but important diagnosis to consider.

A solitary cecal diverticulum is considered to be congenital in etiology and a true diverticulum, defined as a projection including the mucosa, submucosa, and muscularis propria. These projections may arise as early as the 6th week of embryonic development [3]. This is in comparison to a false diverticulum, defined as herniation without muscularis propria, typically seen in adults. False diverticula are generally located at weak points along the colonic wall and are associated with muscle atrophy at the location of penetrating vasa recta [4]. Of interest, Sigaloff and others present four pediatric cases where a solitary cecal diverticulum was false on final pathology, demonstrating that it may be possible to acquire a single false diverticulum as early as the adolescent years [11,13,14]. Other forms of congenital diverticula include Meckel's and appendiceal diverticula, both of which can develop complications including diverticulitis with right lower quadrant pain [5,6] and should be on the differential if appropriate. Overall, right sided diverticular disease should be considered distinct from left sided and is more likely to have a genetic predisposition [4]. These include connective tissue diseases such as Marfan syndrome or Ehlers-Danlos, Williams-Beuren syndrome, or neural abnormalities such as hypo-ganglionosis or aganglionosis of the intestine [2].

2.1. Review of case reports in the literature

A review of literature includes 9 cases of pediatric cecal diverticulitis (Table 1) [10-16]. PubMed was used and search terms included "cecal diverticulitis" AND "pediatric." Some earlier reports were found directly from references of other literature reviews. Ages ranged from 3 to 15-years-old, with median age of 13-years-old. Of those where nationality was specified, 5 were of Asian descent. All 9 cases presented with right lower quadrant pain, with 3 having referred pain to the epigastric or periumbilical regions. The presence of pain ranged from less than 24 h to 3 days before presentation to medical care. Six cases reported leukocyte levels and these were elevated with a range of 12.5–22 k/cumm. A large predominance of neutrophils was also noted and ranged from 72 to 83%.

Ultrasound was the most common initial imaging modality while CT was performed first in 3 cases and used second to confirm US findings in 2 other cases. Ultimately, 4 of the 9 patients were correctly diagnosed pre-treatment with cecal diverticulitis while 5 were diagnosed with or were not able to rule out acute appendicitis. Seven of the 9 reported cases underwent surgical treatment with 6 cases performing ileocecectomy with primary anastomosis, while only 1 case underwent diverticulectomy and appendectomy. The 2 cases that did not have surgery had correct diagnoses of uncomplicated diverticulitis and were initially treated with IV antibiotics. One of these had recurrence with subsequent colonoscopy and endoscopic removal of fecalith.

When compared to the existing literature for adults with cecal diverticulitis, the symptoms are very similar to the cases of cecal diverticulitis in children. One retrospective study found that the pain was more often characterized as dull and lasted for a longer duration, and was not associated with nausea and vomiting as compared to acute appendicitis [3]. However, in another study the correct diagnosis of right sided diverticulitis or possible diverticulitis in adults was still made only 39.4% of the time prior to operation or other treatment. And this study was significantly better than most other studies it compared to, where misdiagnosis occurred over 90% of the time [7]. In regard to laboratory results, some studies have stated that there may be a mildly elevated WBC count with a higher percentage of lymphocytes in acute diverticulitis as compared to what is expected in appendicitis [8]. CT of the abdomen and pelvis with oral and IV contrast is the current test of choice for diagnosing acute diverticulitis, as well as differentiating other conditions that may mimic acute appendicitis [2]. If further imaging is needed to confirm the diagnosis, MRE can be performed. Colonoscopy can be performed in order to rule out Crohn's disease which can show similar inflammatory changes of the colon on CT imaging, but a diverticulum may not be visualized if the opening of the pouch is narrow.

2.2. Management

For treatment of uncomplicated diverticulitis (without phlegmon, abscess, perforation, or fistula [1]) conservative medical management with IV antibiotics is preferred in both children and adults. Surgery is reserved for those with complicated diverticulitis, those that are hemodynamically unstable, or for recurrence of disease [9]. However, because there is such a high rate of preoperative diagnosis of acute appendicitis, surgery often occurs without this determination. Laparoscopic diverticulectomy with appendectomy is the

preferred surgical approach in children and adults. Appendectomy should be performed at the same time in order to avoid misdiagnosis if right lower quadrant pain were to recur in the future [9]. Although a laparoscopic approach is favored, should the lesion not be identified then an open approach is warranted. In the growing child, ileocecectomy should be avoided due to the risk of malabsorption, however, it may be considered based upon the extent of inflammation and infection and if there is perforation involving the colonic wall [15].

3. Conclusion

The most common presentation of right-sided diverticulitis in the pediatric patient is right lower quadrant pain that may or may not refer to the periumbilical region. This condition commonly mimics acute appendicitis, often sending patients to the operating room despite current recommendations for treatment of uncomplicated diverticulitis with conservative medical management. Laboratory findings may be nonspecific or include only a mild leukocytosis and CT is currently the most sensitive and specific imaging modality. When surgery is indicated for complicated diverticulitis or performed to reduce recurrence, we recommend diverticulectomy with appendectomy as a viable and safe alternative over ileocecectomy. As this condition becomes relatively more common in younger and Western populations, it may be beneficial for the pediatric surgeon to be aware of this differential when considering a diagnosis for right lower quadrant pain.

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Fig. 1.

CT of the abdomen and pelvis with IV contrast. Axial image (A) demonstrated cecal diverticulum (black arrows) and coronal image (B) demonstrated thickening and mural edema of the diverticulum (white arrowheads), indicating inflammation.

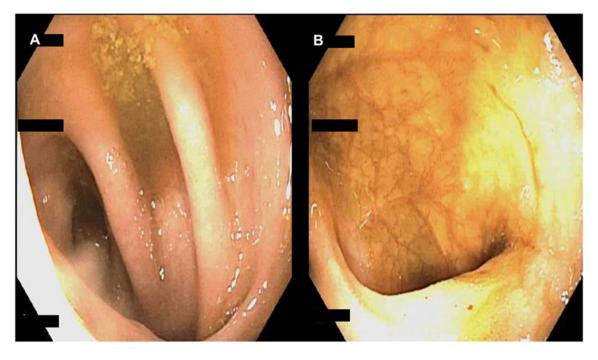


Fig. 2.

Pediatric colonoscopy images of (A) cecum and (B) terminal ileum. Colonoscopy showed that the terminal ileum and entire examined colon appeared normal.

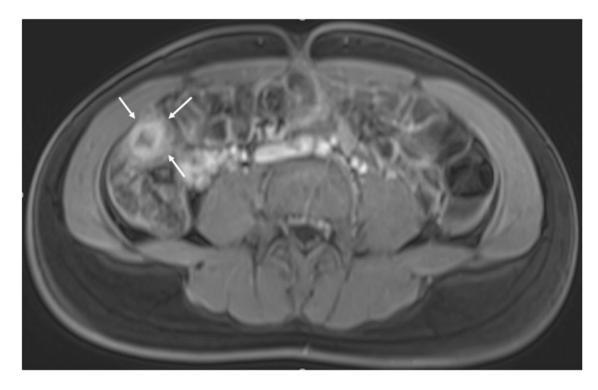


Fig. 3.

Magnetic Resonance Enterography. Axial T1-weighted image acquired after the administration of IV contrast demonstrated mucosal hyperenhancement of the diverticulum (white arrows) with redemonstration of the thickening and mural edema.



Fig. 4.

Intraoperative visual of the anteromedial cecal diverticulum prior to diverticulectomy and appendectomy. Tip of hemostat points to the diverticulum, the appendix appeared normal.

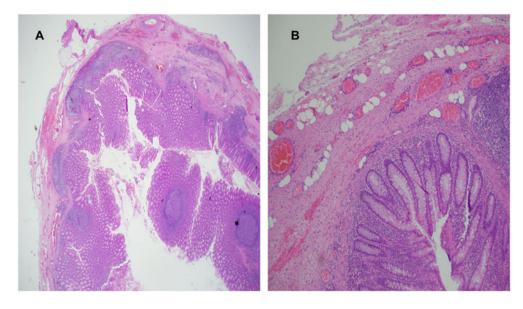


Fig. 5.

H&E stain of cecal diverticulum. Final pathologic diagnosis included portion of bowel with blind end (A) 2x magnification, and (B) 10x magnification, compatible with clinical impression of cecal diverticulum.

	Demographics	Clinical presentation	Positive labs	Pre-op diagnosis and imaging	Treatment	Post-op management	Final pathology
Sigaloff et al., 2005	13-year-old Korean male	2 days of intermittent, worsening RLQ pain that initially referred to periumbilical region	WBC 22000, CRP 24	US – acute appendicitis	Laparotomy and ileocecectomy	IV antibiotics and discharged 6 days post-op	False diverticulum
Bogue et al., 2008	13-year-old Asian male	24 h of RLQ pain	Temp 38 C	US- ascending colonic diverticulitis, confirmed with CT	IV antibiotics	No recurrence of symptoms in 15 months of follow up	N/a
Cheng et al., 2012 #1	15-year-old Hispanic female	I day of epigastric pain localizing to the RLQ with subjective fevers	Temp 38 C, WBC 15000 78% neutrophils	US- free fluid. CT- irregular, asymmetric thickening of the cecal wall and normal appearing appendix, not possible to exclude acute appendicitis	Ileocecectomy with primary end-to-end anastomosis	7 days of IV pip/tazo and discharged 7 days post-op	False
Cheng et al., 2012 #2	3-year-old Caucasian female	3 days of worsening periumbilical pain with RLQ tenderness to palpation	WBC 22600, CRP 15.89	CT- retrocecal abscess containing hyperdense structure	Ileocecectomy with primary anastomosis	7 days of IV pip/tazo and discharged 7 days post-op	False
Rich et al., 2012	10-year-old female	1 day of worsening abd pain with RLQ tenderness to palpation	WBC 12500 72% neutrophils	CT- cecal diverticulitis	Ileocecetomy with primary anastomosis	Discharged 6 days post- op	False
Huntington et al., 2016	9-year-old male	Less than 24 h RLQ pain with nausea and vomiting	WBC 14300 83% neutrophils	CT- lamellated appendicolith, gas distal to obstructing stone, ruptured appendix	Laparoscopic appendectomy converted to open ileocecetomy with stapled ileocolonic anastomosis	Discharged 5 days post- op	True
Yano et al., 2019 #1	11-year-old Japanese female	RLQ pain		US- acute appendicitis with fecalith	Laparascopic converted to laparotomy ileocecal resection and hand-sewn anastomosis	Discharged 7 days post- op	True
Yano et al., 2019 #2	14-year-old Japanese male	RLQ pain and tarry stool		US- cecal diverticulitis with fecalith	Primary: IV antibiotics Recurrence: IV antibiotics, colonoscopy and removal of fecalith with endoscopic forceps	Readmitted 4 days after discharge	N/a
Cil et al., 2019	14-year-old Turkish male	1 day of abdominal pain, nausea, and subjective fevers. Similar but lesser symptoms experienced 1 week ago	WBC 15390 77.3% neutrophils	US- acute appendicitis	Diverticulectomy and appendectomy	Discharged 5 days post- op, subcutaneous fat necrosis resolved within a week	True

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Table 1