

Solitary cecal diverticulitis in a pediatric case: A challenging diagnosis

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Pediatric diverticulosis is a rare pathology, as it is usually described more frequently in the population over 50 years of age in the Western world, being more frequent in men. Its presentation at a younger age is associated with an increased risk of complications, including diverticulitis.^[1] Potier first described cecal diverticula in 1912.^[2] An incidence between 1:50 and 1:300 of appendicitis has been reported, with solitary cecal diverticulum (SCD) having an incidence of 1 to 2% in North America; while 43 to 50% of all cases of colonic diverticulosis are caused by SCD in countries in the Asia.^[3] The exact etiology of this pathology is still unknown; however, some theories have been described about the formation of diverticula.^[4]

As less than 5% of all cases of diverticulosis develop diverticulitis, the initial presentation of a diverticulum with inflammation or infection is extremely rare. Although it has been described that 65% of the population of Western countries have diverticulosis around the age of 85, this pathology is present in only 10% of the population under 40 years of age.^[5] In Western countries, most patients have diverticulitis in the left colon, while in countries such as Asia, the most frequent presentation is on the right side, representing more

Abstract

Pediatric diverticulosis is a rare pathology. In Western countries, most cases of diverticulitis occur in the left colon. In contrast, the rate of right-sided diverticulitis is much more common in Asia. Complicated diverticulitis is extremely rare in children and adolescents. The diagnosis of diverticulitis in pediatrics can be made during the evaluation of abdominal pain or during surgical treatment of the acute abdomen. A six-year-old female patient was admitted with intraoperatively diagnosed cecal diverticulitis. Her medical history was unremarkable. She presented with abdominal pain located in the right iliac fossa of 48 h of evolution, without other symptoms. She underwent surgery for suspected acute appendicitis, finding a normal subcecal appendix and an additional retrocecal plastron-like structure which was initially classified as an appendicular duplication. Plastron release, appendectomy, and accessory structure excision were performed. The histopathological result was reported as cecal diverticulitis. At one month of follow-up, she had no complications. In conclusion, cecal diverticulitis in pediatrics is a rare pathology, difficult to diagnose, and is often confused with acute appendicitis due to clinical findings and complementary studies. Therefore, it is usually diagnosed during examination by laparoscopy or laparotomy.

Keywords: Acute abdomen, cecal diverticulitis, diverticulosis, pediatrics.

than 70% of cases, and lower recurrence rates have been described. In addition, approximately 80% of cases correspond to DCS.^[4,6]

It has been reported that approximately 2 to 9% of pediatric patients with diverticulosis have complicated diverticulitis, which is extremely rare; therefore, to date, only isolated case reports have been made in the literature.^[4,6] The diagnosis of diverticula in children and adolescents usually occurs during the evaluation of abdominal pain or during the surgical treatment of an acute abdomen, occurring in about 1% of cases with a previous diagnosis of acute appendicitis, thereby making a preoperative diagnosis is quite complicated.^[6]

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In this article, we present a case of SCD in a pediatric patient.

CASE REPORT

A six-year-old female patient presented with no significant prenatal, natal or postnatal history, no previous hospitalizations or surgical interventions, with a complete vaccination schedule and a family history of diabetes in her maternal grandfather. She was admitted to the emergency room with a history of continuous abdominal pain located in the right iliac fossa during the last 48 h, which progressively increased in intensity, without being accompanied by fever, vomiting, diarrhea or hyporexia. On physical examination, she presented positive appendicular signs (McBurney, Blumberg,

Psoas, Dunphy). Complementary studies showed normal leukocytes, no neutrophilia, no anemia, normal platelets, normal renal function, normal coagulation times, C-reactive protein of 1.92 mg/dL (reference range: 0.00 to 0.50 mg/dL).

An abdominal ultrasound was performed, which reported the presence of an appendicular plastron. As an acute inflammatory abdomen was presented, suspecting acute appendicitis, exploratory laparoscopy was performed, evidencing the following surgical findings (Figures 1, 2, and 3): subcecal appendix without inflammatory changes of 6×0.5 cm with a good quality appendicular base; a retrocecal structure indicating a second appendix of approximately 1.5 cm similar to appendiceal plastron surrounded by the greater omentum,

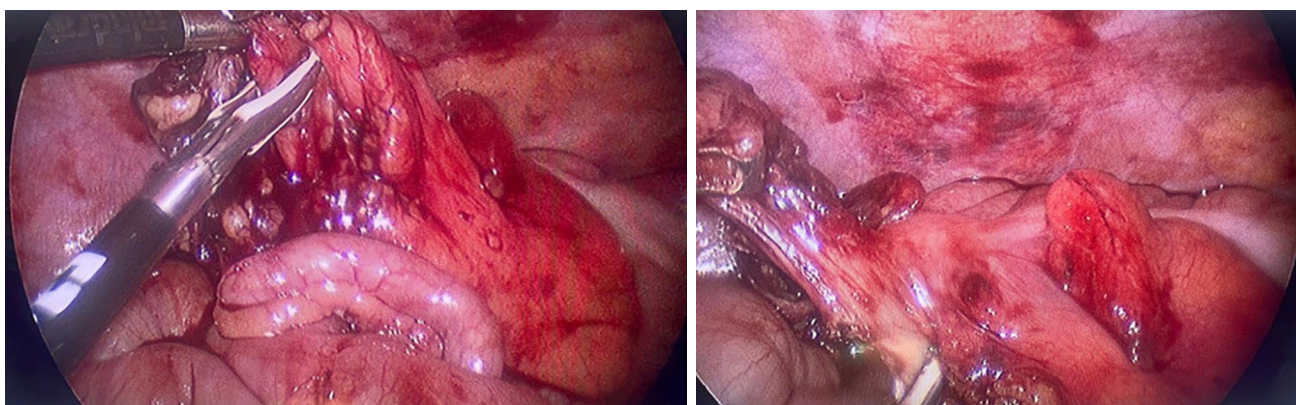


Figure 1. Cecal diverticulum: Release of cecal diverticulum from plastron with greater omentum, abdominal wall, ascending colon and cecum.

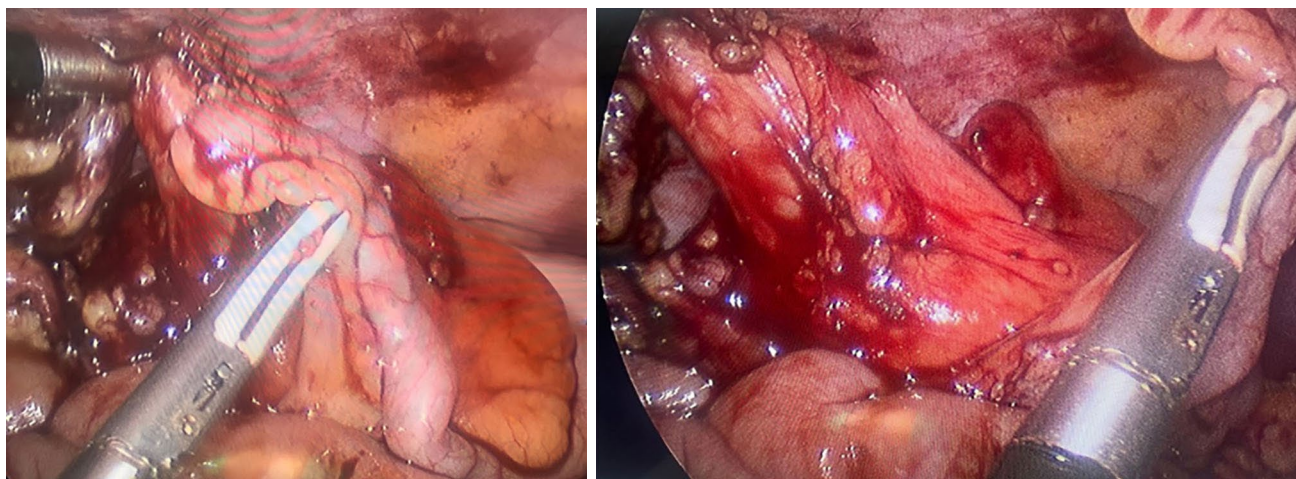


Figure 2. Intraoperative image: On the left, cecal diverticulum, on the right, cecal appendix.



Figure 3. Macroscopic piece: Subcecal appendix on the left, retrocecal structure on the right.

abdominal wall, ascending colon and cecum, which after its release showed signs of necrosis; little free inflammatory fluid in the pouch of Douglas. The rest of the intestinal loops and colon were examined with no evidence of perforations; no Meckel's diverticulum was evident. Plastron release, appendectomy and excision of accessory structure were performed without complications, with a surgical time of 110 min and no significant bleeding.

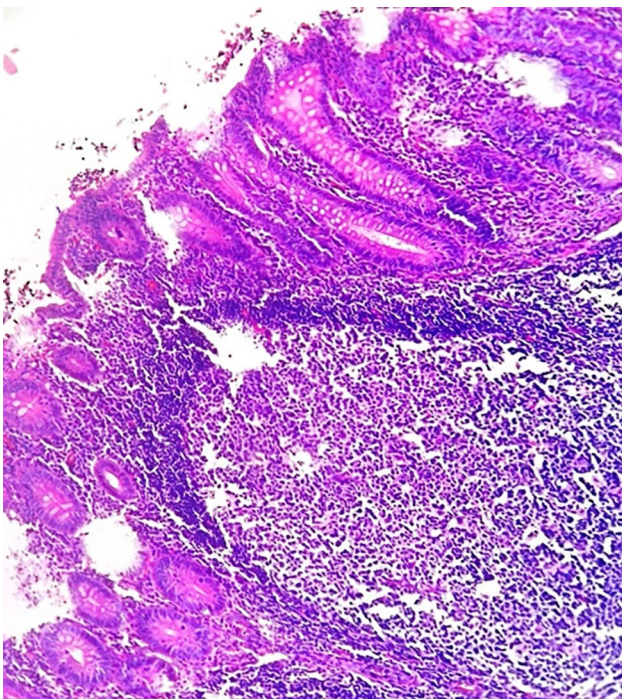


Figure 4. Cecal appendix with adequate preservation of its architecture and serous follicular hyperplasia and periappendiceal adipose tissue with neovascularization, dispersed lymphocytic inflammatory infiltrate, ectasia and vascular congestion.

The patient was admitted to the hospital for postoperative management, receiving antibiotic therapy based on ampicillin plus sulbactam for three intravenous doses, analgesia based on paracetamol with schedule and ketorolac for necessary reasons. Enteral feeding was started 6 h after surgery with clear liquids with adequate tolerance, progressing the diet to soft foods diet. She was discharged 36 h after surgery without complications and without requiring outpatient antibiotic therapy. During her follow-up in the outpatient clinic, the histopathological result was reported as reactive follicular hyperplasia-periappendicitis which was compatible with SCD. At one month of follow-up, the patient had no complications. A written informed consent was obtained from the parents of the patient.

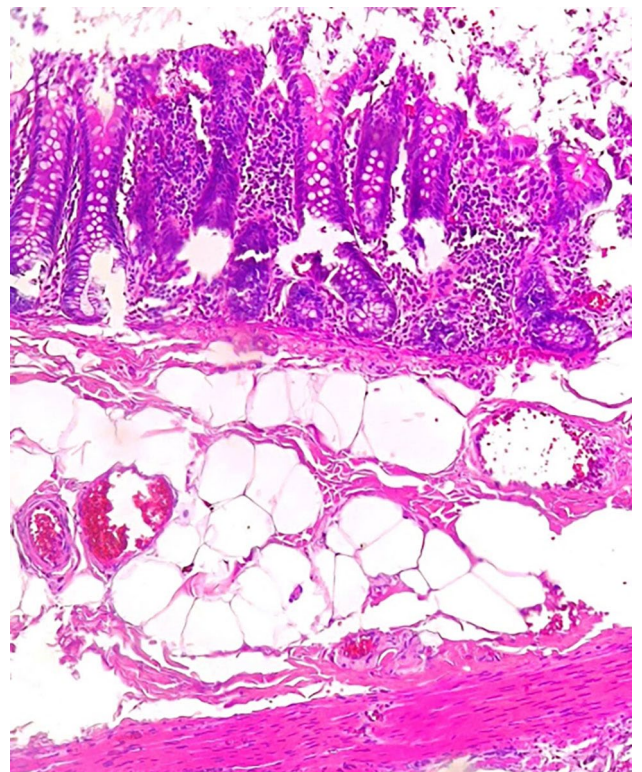


Figure 5. Accessory structure: Fibroconnective and adipose fragments with necrosis, hemorrhage, neovascularization, vascular congestion and lymphocytic and polymorphonuclear inflammatory infiltrate; focally, fragments of the wall of the large intestine with its four preserved histological layers, which is continuous with the aforementioned adipose tissue.

TABLE 1
Solitary cecal diverticulum and its treatment

Grade	Treatment
1: Inflamed diverticulum	Treatment is conservative if the diagnosis is made preoperatively, treatment is appendectomy ± diverticulectomy if the diagnosis is made intraoperatively.
2: Inflamed mass	Treatment is conservative if the diagnosis is preoperative; if the diagnosis is intraoperative, treatment is limited ileostomy or right hemicolectomy.
3: Localized abscess/fistula	
4: Perforation/ruptured abscess with generalized peritonitis	Regardless of whether the diagnosis is preoperative or intraoperative, treatment is limited ileostomy or right hemicolectomy. ^[2,9]

DISCUSSION

Although the exact etiology of cecal diverticulosis is still unknown, there are some theories regarding the causes of diverticula formation, including genetics, inflammation, microbiome, and intestinal motility. The first theory indicates that it is caused by an increase in pressure in areas of the intestine where the walls are weaker; the genetic theory describes that the heritability effect is estimated at 40%, while the non-shared environmental effect is 60%; the motility theory suggests that diverticulosis can result from neural degradation of the myenteric plexus, resulting in uncoordinated contractions and a subsequent increase in pressure on the colon wall.^[4]

Diverticula can be classified as congenital (true) and acquired (false). Regarding the first one, it has been described that it develops due to a congenital defect from the sixth week of gestation of an external pouch of the cecum that involves the three layers of the colon wall (mucosa, submucosa and muscularis propria), while false or acquired diverticula develop from a protrusion of the mucosa and submucosa due to a chronic increase in intraluminal pressure in areas of weakness of the muscular layer. There are modifiable risk factors that, as age increases, will increase the incidence of this pathology: obesity, smoking, a diet low in fiber, diets rich in red meat and fat, and the use of nonsteroidal anti-inflammatory drugs, corticosteroids and opiates. Protective factors include a diet rich in fiber, physical activity and medications (calcium channel blockers, statins and vitamin D). Therefore, a diverticulum of congenital etiology, which can present complications at a relatively early age, is an extremely rare pathology.^[4,5,7]

Cecal diverticulitis can also be classified as solitary, which are usually congenital, and multiple, which are acquired.^[7] In the literature, it is described that between 50 and 60% of the cecal diverticulosis are found in the anterior wall of the cecum, while the remaining cases are located in the posterior wall. In addition, in the majority of cases (80%) these are located 2.5 cm from the ileocecal valve.^[4,7] Left-sided diverticulosis is different from right-sided diverticulosis, as the latter is more likely to be secondary to a genetic predisposition and includes connective tissue diseases such as Marfan or Ehlers-Danlos syndrome, Williams-Beuren syndrome, or neuronal abnormalities such as hypoganglionosis or aganglionosis of the intestine.^[5]

Solitary cecal diverticulum is usually asymptomatic; in approximately 13% of patients, its most frequent complication is diverticulitis, which is often confused with acute appendicitis due to its clinical manifestations and imaging findings. Patients may present with a variety of symptoms such as abdominal pain, bleeding in the stool, and signs of peritonitis. Diverticulosis may manifest as severe chronic abdominal pain or as mild intermittent pain, which is later accompanied by fever, nausea that may lead to vomiting, diarrhea, or constipation. If it evolves into peritonitis, abdominal rigidity, muscle guarding, and the positive rebound sign may be evident. Blood tests may show leukocytosis and elevated inflammatory biomarkers.^[4,6]

The diagnosis of diverticulosis or diverticulitis in a child or adolescent can be carried out during the evaluation of abdominal pain during surgical treatment of the acute abdomen; thus, the true incidence of these pathologies remains uncertain. The preoperative clinical diagnosis is

quite complicated, since this entity is detected in approximately 1% of cases or up to 1/300 cases on with a preoperative diagnosis of acute appendicitis are done. In the study conducted by Uhe et al.,^[6] 67% of patients presented abdominal pain located in the right iliac fossa in 93.2% of them, and 70% of patients underwent exploratory surgery with a preoperative diagnosis of acute appendicitis.^[6]

Abdominal ultrasound and computed tomography (CT) are imaging studies which can be useful for diagnosis. The ultrasound is more accessible and less expensive and has a sensitivity of 91.3% and specificity of 99.8% for diagnosing right-sided diverticulitis, where findings may show “hypoechoic structures of a round or oval shape that protrude from the colon wall, unlike appendicitis, which is usually tubular and may be surrounded by hyperechoic heterogeneous soft tissue that corresponds to inflammation of the pericolic fat.”^[6,8,9]

The management of cecal diverticulitis will depend on the inflammatory state of the lesion and whether or not there is a complication. There are four degrees of diverticulitis according to the management guidelines, in accordance with the recommendations of the American Society of Colon and Rectal Surgeons.^[3,10]

Laparoscopic surgery is recommended, as it is associated with less bleeding, shorter hospital stays and faster recovery. However, it is not always possible as it depends on the location of the DCS, as it is easier to manage the one that originates in the anterior wall than the one that is in the lateral or posterior wall, particularly if it is associated with phlegmon; in these cases, a prolonged dissection may be necessary, or even conversion to open surgery.^[6,7]

In conclusion, cecal diverticulitis in pediatrics is an extremely rare pathology, difficult to diagnose, which is often confused with acute appendicitis due to clinical findings and complementary studies; therefore, it is usually diagnosed during a laparoscopic or laparotomy examination. Treatment depends on the degree of diverticulitis. In the case presented, the diagnosis was made intraoperatively

due to the clinical picture of acute appendicitis, and it was managed by diverticulectomy without complications.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept, control/supervision, data collection and/or processing: F.A.A.V.; Design, analysis and/or interpretation: G.E.T.E.; Literature review, writing the article, references and fundings: G.E.T.E., G.D.S.S.; Critical review, materials: F.A.A.V., G.E.T.E., G.D.S.S.

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