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A rare disease presenting with acute abdominal pain in a girl: Solitary cecal diverticulum

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ABSTRACT

Solitary cecal diverticulum (SCD) is a rare gastrointestinal disease, especially in children, predominantly observed in middle-aged Asian men, and presented with nonspecific symptoms similar to acute appendicitis. Herein we report a case of SCD in a 15-year-old girl who had acute abdominal pain symptoms, and was diagnosed intraoperatively. The SCD, which contained a fecalith, was excised, followed by cecal repair and an appendectomy. Histopathological examination confirmed the diagnosis of SCD. This case highlights the importance of considering SCD in the differential diagnosis of acute abdominal pain, particularly in pediatric patients.

Keywords: Solitary cecal diverticulum, acute abdominal pain, diverticulitis, fecalith

Solitary cecal diverticulum (SCD), first described by Potier in 1912, is a rare gastrointestinal disease (1). SCD has been reported to constitute only 3.6% of all colonic diverticula (2). It has been reported that the disease typically manifests in the fifth decade of life and is more commonly observed in Asian men. SCD often presents with nonspecific symptoms such as right lower quadrant abdominal pain, vomiting, and loss of appetite, making its differentiation from acute appendicitis challenging (3). It is rarely seen in the pediatric population, with only a limited number of case series or reports available in the literature (4-7). In this study, we present a case of SCD in an adolescent girl who presented with acute abdominal pain due to fecalith-induced inflammation.

CASE PRESENTATION

A 15-year-old female patient with no previous medical history was admitted to the emergency department with the sudden onset of abdominal pain and vomiting persisting for two days. Vital signs were within normal limits. Physical examination revealed tenderness, guarding, and rebound tenderness in the right lower quadrant of the abdomen. Laboratory results showed a white blood cell count of 11,510×10^3/mL, a left shift (Neutrophils: 74.3%), and elevated C-reactive protein (17.4 mg/L). An upright abdominal X-ray did not reveal any diagnostic findings. Abdominal ultrasonography (USG) demonstrated edema in the ileocecal loop and adjacent mesentery in the right lower quad-

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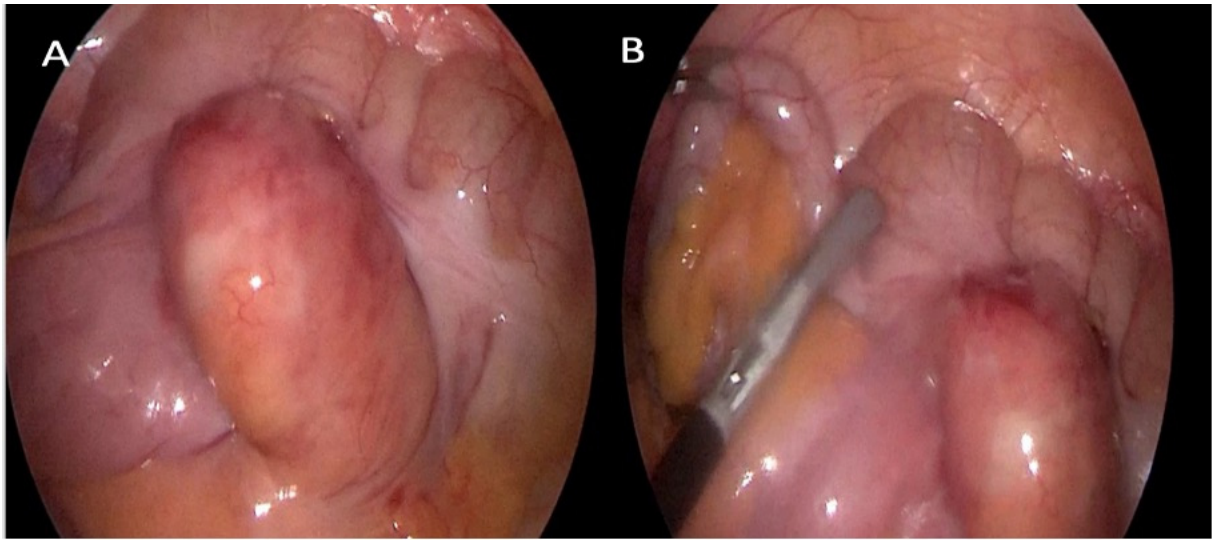

Fig. 1. A) Inflamed mass (SCD) located on the anterior wall of the cecum adjacent to the ileocecal junction during laparoscopic examination. B) Normal appearance of the appendix.

rant, along with approximately 1 cm of free fluid in the Douglas pouch. In addition, a blind-ending luminal structure containing a fecalith was observed on the anterior surface of the cecum. After initial resuscitation, laparoscopy was performed. On laparoscopy, an inflamed, pedunculated mass was identified on the anterior wall of the cecum, while the appendix appeared normal (Fig. 1A and Fig. 1B). When the mass was excised from its neck, it was noted to be a luminal structure containing a fecalith. To ensure safer repair, conversion to laparotomy was performed. Diverticulectomy and bowel wall repair in two layers were

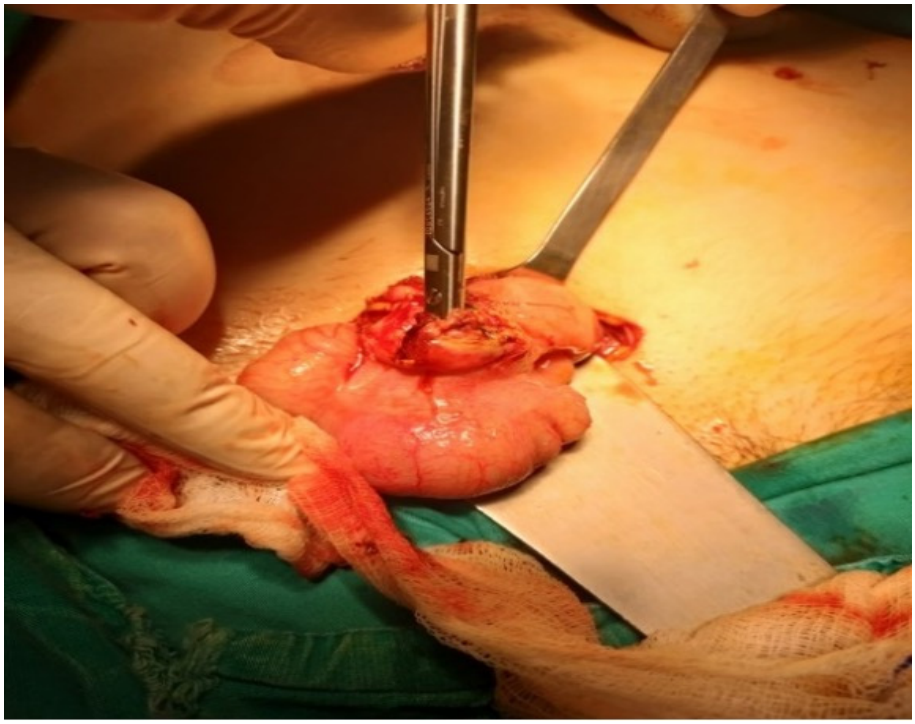

Fig. 2. The connection between the SCD and the cecal lumen is observed during laparotomy.

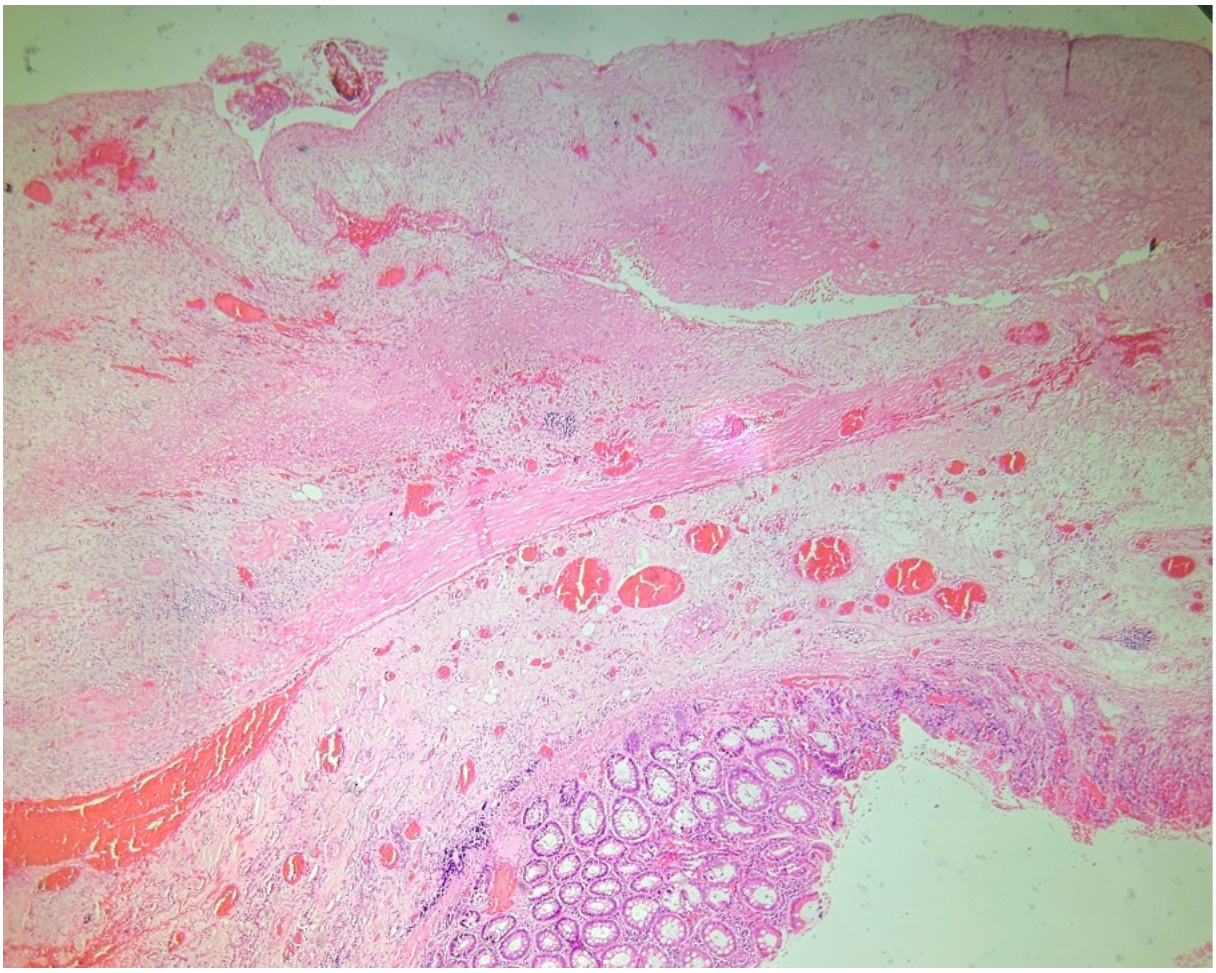

Fig. 3. Histopathologic examination revealed that the SCD structure was lined with colonic epithelium and involved all layers of the bowel, exhibiting vascular dilation, congestion, and acute inflammation within its wall (Hematoxylin-eosin, ×40).

performed (Fig. 2). The procedure was concluded after formal appendectomy. The postoperative course was uneventful. Histopathologic examination revealed colonic tissue involving all bowel layers, exhibiting vascular dilation, congestion, and acute inflammation, and suggested an SCD association with diverticulitis (Fig. 3). The patient was followed up for two years without complications.

DISCUSSION

Colonic diverticula are commonly observed in older age groups and are rarely symptomatic. The etiology of the condition has been linked to a sedentary lifestyle, obesity, and a diet deficient in fiber. It has been suggested that diverticula are more frequently observed in the left colon due to increased intraluminal pressure, and the disease is considered an acquired pathology (8). On the other hand, the incidence of right colonic diverticula has been reported to increase in conditions such as connective tissue disorders (e.g., Marfan syndrome, Ehlers-Danlos syndrome, Williams-Beuren syndrome) and neuroenteric anomalies like intestinal hypoganglionosis (9). These types of diverticula may be located throughout the colon and may be numerous. Because they are contained within the mucosa-submucosa without involvement of the muscularis layer, it is still debated whether they are true diverticula. SCD is considered distinct from other colonic diverticula due to its location and histopathologic properties, which involve all bowel layers (10). One theory proposes that SCD arises from a persistent "temporary appendix" structure, which develops dur-

ing the first six weeks of the embryologic period and fails to regress as expected. SCDs are typically located approximately 2-3 cm from the ileocecal valve and are positioned anteriorly (11). In our case, the diverticulum was found on the anterior surface of the cecum, attached via a narrow neck similar to the appendix. The histopathological examination revealed that the lesion involved all layers of the normal colonic wall, and we believe this supports the congenital malformation theory of SCD etiology mentioned above.

Since its first description, up to 1,000 cases of SCD have been reported in the literature across various age groups (12). In a study by Sardi *et al*. (2), which included 881 adult cases, SCD constituted 3.6% of all colonic diverticula, with an average age of 43.6 years. It was noted that colonic diverticula are more frequently encountered in men compared to women. In a systematic review, data were collected from a total of 988 patients, aged 20 to 73 years. The male-to-female ratio in this study was reported to be approximately 1.12:1 (13). In the pediatric population, it is difficult to estimate the incidence and the male-to-female ratio due to its rarity. To date, 18 cases have been reported, with most of them being case reports (4-7).

SCDs remain asymptomatic unless complicated, and most of them can be detected incidentally during surgery. Symptomatic cases most commonly present with acute abdominal symptoms due to diverticulitis, bleeding, or perforation (14). Diagnosis of SCD, especially distinguishing it from appendicitis, can be difficult (2). In a systematic review of adult series, the most common symptoms of SCD were right lower quadrant abdominal pain, nausea and/or vomiting, and fever, with frequencies of 93.2%, 35.4%, and 26.9%, respectively (13). In our case, the main complaints were acute abdominal pain, nausea, vomiting, and loss of appetite, which mimicked acute appendicitis. Even in symptomatic cases, preoperative diagnosis of SCD through imaging is challenging (15).

In the literature, 22.8% of SCD patients were diagnosed via radiological imaging, while the remainder were diagnosed during surgery (13). In a study of 19 patients diagnosed with SCD, Wyble *et al*. (14) reported that only 2 (11%) were diagnosed preoperatively with detailed imaging, but these cases had previously undergone appendectomy, which allowed for the diagnosis to be made. On the other hand, in a prospective study by Chou *et al*. (16), involving 934 patients presenting with right lower quadrant pain, it was emphasized that USG is a more effective technique for differentiating SCD from appendicitis, with a sensitivity of 91.3% and specificity of 99.5%. In studies involving adults, the detection of a segmentally thickened colonic wall with an oval, hypoechoic structure protruding from the right colon on US has been suggested as an indicator of uncomplicated right colonic diverticula (17). Computerized tomography (CT) has also been used to diagnose SCD. If complicated, some characteristic appearances, such as thickening of the right colon wall, inflammation in the surrounding fat tissue, abscess formation, and local free air, may be diagnostic. However, these nonspecific imaging findings may also be seen in other pathologies, particularly in cecal carcinoma (18).

In pediatric SCD cases and series, no typical findings have been reported, and it is often incidentally discovered during surgery for acute abdominal conditions, especially appendicitis (4-7). In our case, preoperative USG suggested appendicitis with a fecalith. As is known, when a fecalith is found on imaging studies, acute appendicitis is typically diagnosed definitively. We also diagnosed our patient with acute appendicitis. This is the second case report of a pediatric patient presenting with diverticulitis caused by fecalith-induced inflammation.

Colonic diverticula, commonly encountered in the adult patient population, have established treatment standards. In uncomplicated cases, a conservative approach involving bowel rest and intravenous antibiotics is preferred, while in complicated and symptomatic cases, more aggressive surgeries such as diverticulectomy, ileocolic resection, and right hemicolectomy are performed (19). On the other hand, the recurrence rates of uncomplicated SCD during follow-up have been reported to range from 0% to 25%. For cases with frequent recurrences or complicated SCD, surgical resection is undoubtedly the most appropriate treatment option (20). The choice of surgical procedure depends on the location of the diverticulum and whether it is complicated. In cases with limited inflammation around the diverticulum, diverticulectomy with primary repair and appendectomy is recommended, while ileocolic resection or right hemicolectomy is recommended for complicated cases (21).

Lane *et al*. (19) have recommended right hemicolectomy as the first-line treatment for adult SCD cases. In their study, the mortality rate for those who underwent right hemicolectomy was found to be 18%, and they concluded that hemicolectomy should be reserved for complicated cases. Diverticulectomy can be performed by laparotomy or laparoscopy depending on experience. Due to technical difficulties, prolonged operative time, and increased anesthesia risks, laparoscopic surgery may not be suitable for complicated cases (22, 23).

Because of pediatric SCD series are limited, there is no established standard of the management of SCD. In children with SCD, it is reported that, instead of aggressive approaches, the less aggressive surgical methods for treatment of SCD are preferred (4-7). In our presented case, due to the limited inflammation around the diverticulum, diverticulectomy was preferred, and laparoscopy was converted to open surgery for safe excision. In seven (39%) of the reported pediatric SCD cases from four studies, conservative treatment was performed as the first-line therapy. Among these patients, conservative management was successful in four cases, while two patients subsequently required surgical intervention. Additionally, in one patient with a fecalith detected within the SCD, diverticulitis was treated by colonoscopic removal of the fecalith (4-7).

Although conservative management of uncomplicated SCD is considered the first-line treatment in adult series, surgery may be mandatory when treatment failure and recurrence occur. In various studies, no recurrences or major complications have been reported during follow-up in patients who underwent either simple diverticulectomy or hemicolectomy (19-23). In our case, no recurrences or complications were observed during the two-year follow-up period.

CONCLUSION

SCD is a rare condition in children. Due to the absence of typical signs and symptoms or imaging characteristics, it is often detected during acute abdominal surgery. There is no standardized approach to its treatment. When symptomatic or incidentally detected, simple excision is a safe and effective treatment for SCD.

Ethical Statement

Ethics Committee approval is not required for this study. This study is a case report.

Patient' Consent

Patient was informed about the purpose of the case report, and written informed consent was obtained from the patient's family for this publication.

Authors' Contribution

Study Conception: HÖ; Study Design: HÖ; Supervision: MK; Funding: M/A; Materials: HÖ, MÖ; Data Collection and/or Processing: HÖ, MÖ; Statistical Analysis and/or Data Interpretation: HÖ, MÖ; Literature Review: HÖ; Manuscript Preparation: HÖ, MÖ, MK; and Critical Review: MK.

Conflict of interest

The authors disclosed no conflict of interest during the preparation or publication of this manuscript.

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Editor's note

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