

CECAL DUPLICATION CYST PRESENTING WITH COMPLICATIONS IN EARLY CHILDHOOD: A CASE REPORT

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Abstract. Alimentary tract duplications are a rare congenital anomaly. Their clinical manifestation may vary depending on the location, size and communication with the adjacent bowel. We present a rare case of a 2-year-old girl who was admitted with intermittent abdominal pain, low-grade fever, and signs suggestive of complicated appendicitis. Laboratory investigations revealed elevated inflammatory markers. Abdominal ultrasound and CT scan demonstrated a right-sided mass compressing the cecum, with radiological features suggestive of a fecaloma, possible intussusception, or retroperitoneal abscess. Surgical exploration revealed a large retroperitoneal mass adherent to the cecum and surrounding structures. An ileocecal resection with end-to-side ileocolic anastomosis was performed. Histopathological analysis confirmed a complicated cecal duplication cyst with fecal impaction and chronic inflammation. Cecal duplication cysts, though rare, should be considered in the differential diagnosis of right lower quadrant masses in children. Surgical resection remains the definitive treatment, particularly in complicated cases. Timely detection and appropriate surgical management can lead to excellent outcomes.

Key words: gastrointestinal duplication, diagnostics, operative treatment, case report

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INTRODUCTION

Alimentary tract duplications, also known as duplication cysts, are rare congenital anomalies [1]. They have an incidence of 1 in 4,500-5,000 births, with slight predominance in males. They are believed to occur between the 4th and the 8th gestation week, although the exact etiology is yet to be determined, with multiple theories being discussed [1]. The most prominent theory is a defect in notochord splitting. Proposed theories are also par-

tial twinning, recanalization defects and the influence of environmental factors [2]. Gastrointestinal duplications are defined by three characteristic features: intestinal smooth muscle wall, intestinal mucosa lining, close proximity to a part of the gastrointestinal tract [3]. According to the form, there are two types of duplications – cystic and tubular. The cystic type usually does not interact with the adjacent bowel, but the tubular type may have one or more interactions [4]. These lesions can be divided into foregut, midgut, and hindgut duplications, depending on which part of

the gastrointestinal tract they are intimately attached to [3]. Duplications of the ileum are most common (31%), colonic duplications account for 13%, while cecal duplications are extremely rare (0.4%) [2]. Gastrointestinal duplications can present as an incidental finding during routine investigations. Their clinical manifestation depends on the size, location and presence of luminal connection. Most duplication cysts present symptomatically within the first two years of life with abdominal distention, intestinal obstruction or gastrointestinal bleeding [4].

We report a rare case of an infant diagnosed with a complicated cecal duplication cyst containing impacted fecal material, successfully managed with bowel resection.

CASE PRESENTATION

A two-year-old girl presented at our center with intermittent abdominal pain and reduced appetite for the

past week, with the pain becoming more and more severe, along with episodes of fever (38.7° C) for the past two days. No vomiting was registered. Stool was normal. The clinical exam demonstrated spontaneous and palpable pain and tenderness in the right abdomen, with negative peritoneal signs. Bloodwork exhibited leukocytosis (15.1 G/L) with marginally low hemoglobin (96 g/L) and elevated serum C-reactive protein (CRP) of 7.02 mg/dL (<0.5 mg/dL norm). The initial abdominal ultrasound showed an intussusception-like structure in the right lower quadrant, with a hyperechogenic mass compressing the cecum (Fig. 1). The latter had markedly thickened walls (6-7 mm).

The abdominal X-ray revealed a heterogenic oval fecaloma-like structure with a diameter of 38 mm in the right half of the abdomen (Fig. 2).

Negative-contrast colon fluoroscopy (part of the diagnostic algorithm for intussusception at our institution) was unremarkable (Fig. 3). Abdominal computed to-

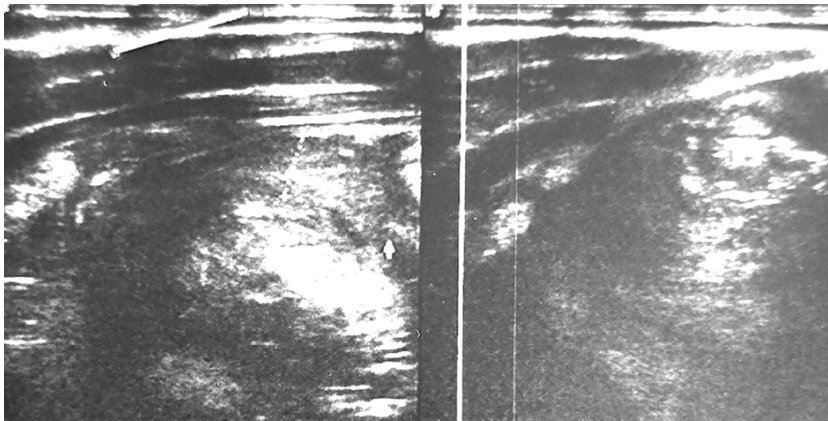


Fig. 1. Abdominal ultrasound showing a hyperechogenic mass compressing the cecum, which had markedly thickened walls (6-7 mm)



Fig. 2. Abdominal X-ray showing a heterogenic oval fecaloma-like structure with a diameter of 38 mm in the right half of the abdomen



Fig. 3. Negative contrast fluoroscopy demonstrating no evidence for intussusception

mography (CT) – scan findings included a large hypodense formation with a hyperdense border at the right lateral canal, inflammatory changes in the cecum and colon wall, retroperitoneal fat and the adjacent sixth hepatic segment (Fig. 4 and Fig. 5).

The findings also suggested an interaction between the formation and the adjacent colon. The appendix was with no pathological findings. Reactive lymphadenomegaly was noted. A fecaloma with stercoral colitis and covered bowel perforation with a retroperitoneal abscess were suggested as most probable diagnoses. Upper and lower GI endoscopies were performed, but the findings were

unremarkable, with the colonoscopy unable to find a fecaloma or (?) along the entirety of the colon (Fig. 6 and Fig. 7).

A decision for surgical exploration was made. Supraumbilical transverse laparotomy discovered a large retroperitoneal mass, contiguous with the posterior wall of the cecum and the ascending colon. The mass was also adherent to the right kidney and sixth hepatic segment. The appendix was anatomically normal. The lymph nodes in the ileocecal region were enlarged. The mass was mobilized, along with the cecum, and the ascending colon. (Fig. 8) An ileocecal resection was reformed an-bloc with the for-

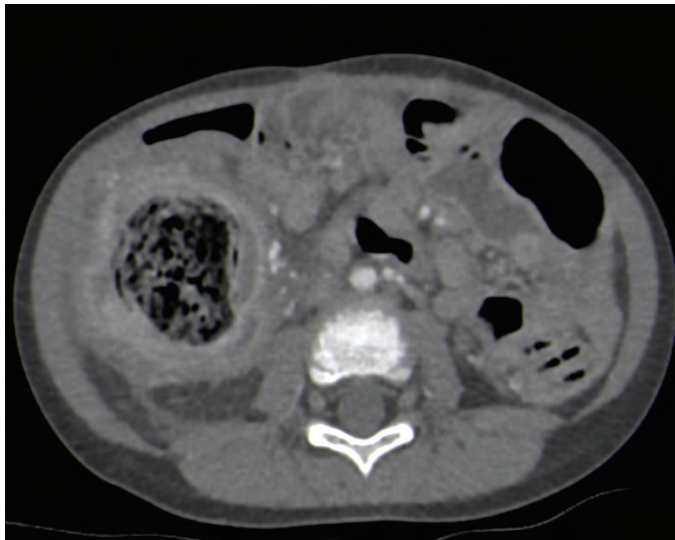


Fig. 4 and 5. Abdominal computed tomography scan demonstrating a large hypodense formation with a hyperdense border at the right lateral canal, inflammatory changes in the adjacent structures

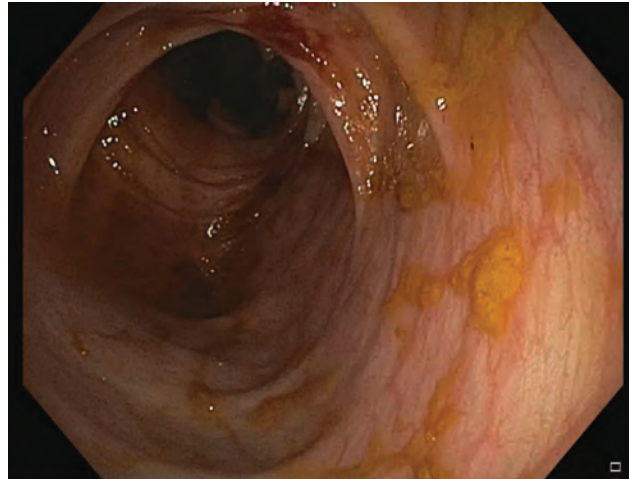
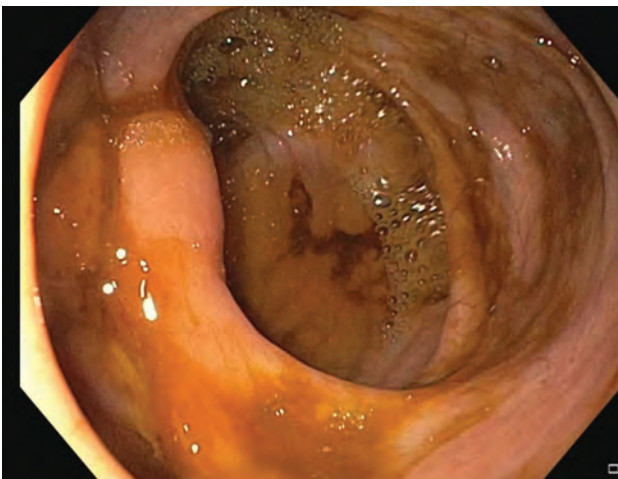


Fig. 6 and 7. Colonoscopy images showing intact cecum and ascending colon

mation (Fig. 9), followed by a terminolateral ileocolic anastomosis. An 18 Fr tube drainage was placed.

The postoperative period was without complications. The histopathological examination of the resected material demonstrated a communicating cecal duplication with fecal impaction and severe inflammatory changes in the wall of the cyst and the adjacent bowel (Fig. 10).

DISCUSSION

Clinically duplication cysts may manifest with palpable mass, gastrointestinal bleeding – in cases where the internal lining consists of ectopic gastric mucosa, characteristics of bowel obstruction – abdominal pain

and distention, vomiting, or even perforation in complicated cases [5]. Malignant transformation can also be observed [6]. Differential diagnosis of gastrointestinal duplications comprises lymphangioma, choledochal cyst, ovarian cyst in females, intussusception, periappendicular abscess [7]. In the vast majority of reported cases cecal duplications in children present with symptoms of bowel obstruction [2, 5, 7-11]. In a retrospective analysis spanning 15 years, Rattan observed that the majority – over 90% – of gastrointestinal duplication cases were associated with subacute intestinal obstruction [12]. In our case the clinical manifestation resembled mostly complicated appendicitis. Dajenah et al. report a case of a 23-year-old patient with cecal duplication cyst presenting as peri-

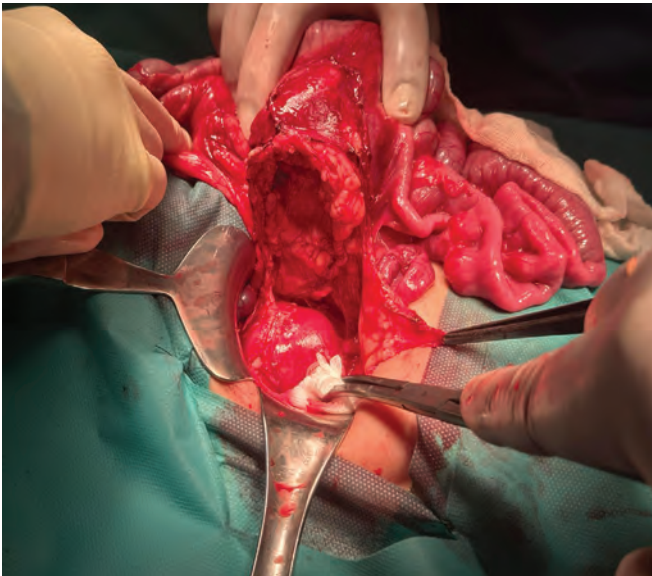


Fig. 8. Intraoperative image of the mobilized cecum and right colon with the formation

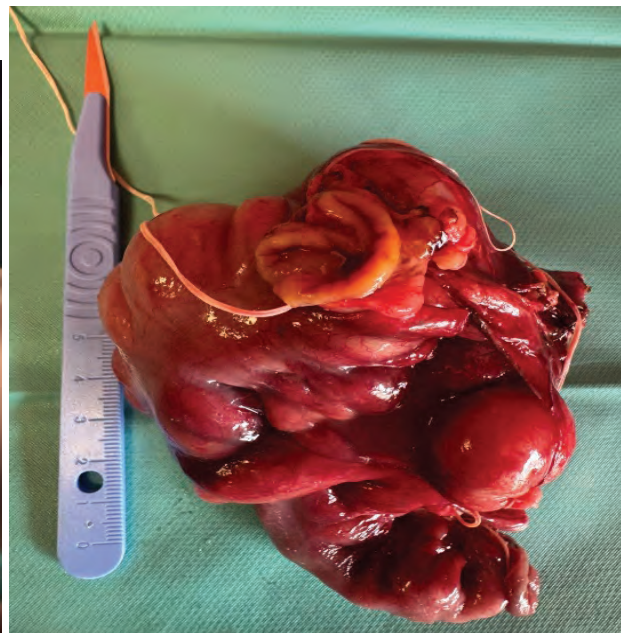


Fig. 9. Postoperative image of the resected specimen

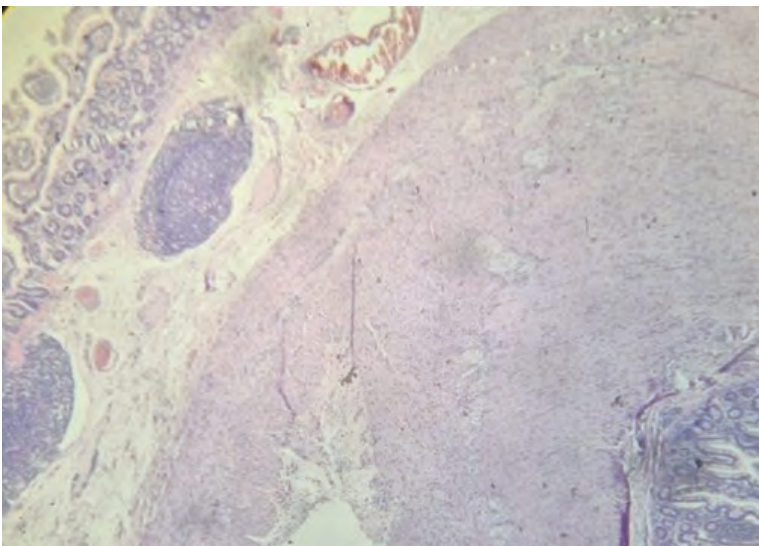


Fig. 10. Hematoxylin-eosin staining (100x) demonstrating layers of the wall of duplication cyst

appendicular abscess [13]. Ahmed et al. presented a 10-month-old child with bowel obstruction, diagnosed with intestinal duplication with fecal impaction [14]. They report tubular colorectal duplication sharing the same wall with the native bowel with no interaction between them [14].

The diagnostic algorithm for this type of pathology may include different imaging modalities according to the presenting symptoms [3]. An increased frequency of prenatal diagnosis of enteric duplications is reported due to prenatal ultrasound. This allows timely management in the neonatal period before complications arise [8]. Gastrointestinal duplications have specific ultrasound characteristics – “double wall” sign – hyper-echoic mucosa surrounded by hypoechoic muscular layers [15]. In our case the ultrasound was suggestive of duplication. The internal contents of the lesion were heterogeneous, differing from the typically hypoechoic appearance seen in cystic duplications. Contrast colonic fluoroscopy can be used to exclude intussusception. Computed tomography (CT) and magnetic resonance imaging (MRI) are employed when more detailed anatomical visualization is required [8]. In the presented case a CT scan was used to delineate whether the symptoms and ultrasound findings were related to periappendicular abscess, intussusception, cecal fecaloma with stercoral colitis, covered bowel perforation or a malignant process. Endoscopic procedures can show outer compression of communication between the duplication and the native bowel. The colonoscopy in our case could not depict an interaction between gut and the duplicated part. This could be explained by the hypothesis that functional closure of the interaction pathway might have happened, as postulated by Kimura et al [16].

The treatment of gastrointestinal duplications is surgical – elective or urgent in complicated cases. The operative approach for abdominal duplication cysts may be conventional or minimally invasive [17]. In the presented case the conventional surgery was deemed optimal due to the inconclusive diagnostic findings. The location and morphological type of duplication can determine the preferred surgical management. Cystic duplications usually are resected along with the native bowel and an anastomosis is performed [8, 12]. In cases involving tubular duplications or cystic duplications of the stomach and duodenum, surgery usually entails excision of the duplication and mucosal stripping [8, 12]. This approach could be used also in cases of duplications of the ileocecal region in order to preserve the ileocecal valve [12, 15]. In the presented case, due to the chronic inflammation, the wall of the cystic duplication could not be delineated from the wall of the caecum. A complete excision of

the formation was performed with ileocecal resection and an end-to-side ileo-colic anastomosis.

CONCLUSIONS

Cecal duplication cysts are rare congenital malformations that may clinically and radiologically resemble more common conditions, such as complicated appendicitis or intussusception. In this case, the unusual presence of fecal material within the duplication led to a challenging preoperative diagnosis. Surgical exploration and histopathological confirmation remain crucial for definitive diagnosis. Complete resection, often including the adjacent bowel, is the treatment of choice. Awareness of this rare entity is essential for pediatric surgeons and radiologists, particularly when dealing with atypical right-sided abdominal masses in infants and young children.

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