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Different clinical presentations, diagnostic difficulties, and management of cecal duplication

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Abstract

Background: Cecal duplications (CDs) are very rare, representing 0.4% of all gastrointestinal duplications. This study evaluates the variable clinical presentations, imaging workup, and surgical management of CDs.

Methods: The records of 7 patients who underwent surgery between April 2001 and December 2011 for CD were retrospectively reviewed. Sex, age, duration of complaints, diagnostic studies, surgical intervention, and pathologic findings were recorded.

Results: The median age was 8 months, and mean age was 1.65 ± 2.88 years. Complaints were abdominal pain, rectal bleeding, vomiting, cough and dyspnea, and a prenatally detected cystic mass. The patients were referred with the diagnosis of appendicular abscess, necrotizing enterocolitis, gastrointestinal lymphoma, and intraabdominal cystic mass. Abdominal distention, signs of peritonitis, substernal retraction, fullness at right lower quadrant, and normal findings were detected on physical examination. Diagnostic imaging included plain abdominal radiography (7), ultrasonography (7), computed tomography (3), and magnetic resonance imaging (2). A cystic mass was reported on radiologic studies in 6 patients and appendicular abscess in 1. Cyst and cecum were resected, ileocolostomy was performed in 6 patients, and cyst excision was performed in 1. The diagnosis of duplication cyst was made by pathologic examination in all patients.

Conclusions: Cecal duplications may be detected incidentally; however, they may mimic appendicular abscess, a tumor mass, or necrotizing enterocolitis. Whether cystic lesions are symptomatic, they should be excised when detected because of possible complications.

Duplications of the alimentary tract are relatively rare developmental anomalies that can occur anywhere from the mouth to the anus. In approximately 80% of patients, the duplication is diagnosed before 2 years of age [1-4]. The small intestine is the most commonly affected site. Colonic duplication occurs in 3% to 20% of cases, whereas cecal duplication (CD) is extremely rare with a reported incidence of 0.4% of all alimentary tract duplications [2-7]. A search of the literature showed that only 31 cases of CD have been reported, indicating its rarity. However, Iyer and Mahour [8]

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reported a series in which 17% of alimentary tract duplications were CDs. Symptoms and findings vary according to location and size of the duplication as well as the presence of ectopic gastric mucosa [2,8,9]. Nonetheless, different and rare clinical presentations have been described [2-4,10]. We present our experience of CD in 7 infants and children with varying clinical presentations.

1. Materials and methods

The study includes 7 patients who underwent surgery between April 2001 and December 2011, with the preoperative diagnosis of intraabdominal mass or acute abdomen, and in whom CD was diagnosed postoperatively. The patients' records were retrospectively reviewed, and sex, age, duration of complaints, diagnostic studies, surgical intervention, and pathologic findings were recorded.

2. Results

The median age was 8 months, and the mean age was 1.65 ± 2.88 years (range, 10 days to 8 years). The male-female ratio was 6:1. In all, 3 of the patients were admitted during the newborn period. Gastrointestinal symptoms were observed in all 3, and respiratory symptoms, in 2. One patient was diagnosed with a cystic abdominal mass during the workup for a possible liver mass, and 1 patient was diagnosed with an intraabdominal cyst on prenatal ultrasound. The patients' characteristics and diagnosis at admission are summarized in the Table. The radiologic studies performed were as follows: plain abdominal radiogram (n = 7), ultrasonography (n = 7), computed tomography (CT) (n = 3), and magnetic resonance imaging (MRI) (n = 2). A gasless area at the cyst location was observed on plain abdominal radiograms in the 3 newborn patients. Imaging studies showed a cystic mass in 6 patients and what was interpreted as an appendicular abscess in 1 patient. Ultrasonography showed thick-walled cysts in 5 patients and intense, particulate cystic contents in 4 patients. A septated cystic appearance was detected by ultrasonography (US) in only 1 patient. Other pathology noted was hepatic solid mass, which was diagnosed as hemangioendothelioma, via liver biopsy in 1 patient. The preoperative diagnoses of the 7 patients are summarized in the Table. A cystic mass attached intimately to the cecum was found in all patients at operation. Cecal resection with the cyst and ileocolostomy was performed in 6 patients. Excision of the cyst alone was performed in only 1 patient who did not have a pronounced shared muscle wall between the cyst and cecum (Fig.). Colonic mucosa was observed at the inner layer of the cyst, in all the patients. Gastric metaplasia was noted in 1 patient. The mean duration of hospitalization was 6.5 ± 1.04 days. The postoperative period was uneventful in all patients.

3. Discussion

Duplication of the alimentary tract is diagnosed based on 3 characteristics, mostly intimate attachment to part of the alimentary tract, presence of a well-developed coat of smooth muscle, and mucosal lining from some part of the alimentary tract [11,12]. Although duplications occur more commonly in females (male-female ratio, 1:2.3), there are some reports that it is more common in males [12-14]. The male-female ratio in the present study was 6:1. In 6 of the patients, there was a common (shared) smooth muscle wall between the cyst and alimentary tract.

Common signs and symptoms of duplication are related to location, type, size, and the presence of ectopic gastric mucosa. Clinical findings in patients with a duplication include recurrent abdominal pain, vomiting, abdominal distention, intestinal obstruction, palpable abdominal mass, failure to thrive, constipation, and occasionally bleeding from ectopic gastric mucosa [5,8,14,15]. Nonetheless, duplication cysts can sometimes be detected incidentally. A cystic mass was detected in 2 patients who presented with respiratory symptoms, in the present study.

Abdominal pain can be related to distention of the cyst, owing to the accumulation of secretions or to cyst compression of the adjacent bowel segment. The causes of abdominal pain were acute abdominal complications from intussusception and infection of the content of the cyst in 2 of our patients. Gastrointestinal bleeding can occur owing to ulceration that develops because of ectopic gastric mucosa in the cyst [15]. Ectopic gastrointestinal mucosa occurs in 30% of all gastrointestinal duplications [3,5]. Although Iyer and Mahour [8] reported ectopic gastrointestinal mucosa in 48% of their patients, only 11% of their patients presented with gastrointestinal bleeding. In the present study, 1 patient had gastric metaplasia.

In particular, colonic duplication can be associated with various congenital anomalies; with genitourinary anomalies being most common [12,15]. None of the patients in the present study had a genitourinary anomaly. Hepatic hemangioendothelioma was observed in 1 patient in the present study, as an accompanying pathology, which has not been reported previously. Intestinal obstruction, volvulus, perforation, intussusception, and malignancy are possible complications of colonic duplication cysts [8,9,16]. Malignant transformation of duplication cysts is very rare in children [3,5,17,16].

Although ultrasonography, CT, and MRI are frequently used preoperatively for diagnostic imaging, the correct preoperative diagnosis is made in less than 25% of cases [3]. Definitive diagnosis of CD is based on surgical observation that the cyst is related to the cecum and