Huge cystic communicating duplication of the right colon with perforated appendicitis

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CASE REPORT

A 15-YEAR-OLD GIRL, a high school student and volleyball player, was referred to our hospital with severe acute abdominal pain and a high fever (up to 38.5°C). We found generalized abdominal tenderness with rebound tenderness and guarding, with the point of maximum tenderness in the upper abdomen above the navel. Computed tomography (CT) depicted a huge distended right colon occupying the abdomen from the upper right quadrant to the entire pelvic region and containing massive solid feces. Because of the distension of the right colon, the appendix was shifted toward the left to the upper abdominal midline above the navel. The swollen appendix contained multiple fecaliths with localized ascites on the appendix (Fig 1). Laboratory values were almost all within normal range, except leukocytosis (16,000/mm³) and C-reactive protein elevation (12.0 mg/dl) were found. Panperitonitis was diagnosed owing to the perforated appendix and severely distended right colon, and we planned an emergency laparotomy under general anesthesia, even though we could not fully determine the reason for solid feces within the dilated right colon. Upon entrance into the abdomen, the perforated appendix, as diagnosed preoperatively, and the apparently normal cecum were identified just under the midline incision. After routine appendectomy, we realized that the lower abdomen was wholly occupied by a cystic mass that communicated with the cecum, and that this mass was a huge cystic communicating duplication of the right colon (35 × 15 × 6 cm, 2,000 g, Fig 2). Resection of the abnormal colon was performed; no other abnormalities, including malrotation, Meckel’s diverticulum, or genitourinary system or vertebral abnormalities were found. The resected specimen contained massive solid paste-like feces; no malignant lesion or ectopic gastric mucosa was identified. The myenteric neural plexus was less apparent on the cystic formation than on the tubular part of the duplication.

DISCUSSION

Duplication of the alimentary tract is a rare congenital anomaly. Occurrence at the ileocecal valve accounts for 30% of cases of alimentary tract duplication, and occurrence at the colon accounts...
Several reports have described patients with colonic duplication who show signs and symptoms consistent with appendicitis, but we could not find a previous case of colonic duplication actually accompanied by perforated appendicitis, with the exception of cases of double appendix. Complications of duplication, such as volvulus, intussusception, small bowel obstruction, and hemorrhage of ectopic gastric mucosa or malignant lesions have been reported. In the present case, the huge feces-filled duplication may have caused multiple fecaliths to develop in the appendix, resulting in appendicitis and perforation. This may be an important finding and complication of distended right colonic duplication.

Similar to other authors who mention the difficulty in diagnosing alimentary duplication before laparotomy, we also were unable to identify the duplication preoperatively. Moreover, the patient required an emergency appendectomy. The CT scan was useful in showing that, because of the severely distended colon, the appendix was located above the navel on the midline. Appendectomy was indicated; however, treatment of the distended feces-filled colon was difficult without a true preoperative diagnosis. Consequently, we thought the best surgical treatment to be resection of the duplication on the basis of symptoms and the potential risk of carcinogenesis and perforation. CT may be important in determining whether a severely distended colon represents a huge cystic duplication, which can occur even in adolescents and adults.

Huge duplications in adults and adolescents have been reported previously, but less is known about the decrease of myenteric neural plexus. In the patient presented in this study, the tubular part of the duplication was innervated by normal myenteric plexus, but the cystic part was less so. Two causes are postulated: the plexus had barely developed on the cystic part, or the apparent decrease of plexus density was caused by secondary distention at the terminal region of the duplication, which was under long-term peristaltic pressure. A previous report exists of an adult without the myenteric

Fig 1. Preoperative CT findings. A, A massive amount of feces was present in the severely distended duplicated right colon. B, The appendix was shifted to the midline of the abdomen and contained calcified fecaliths.
plexus on the huge cystic noncommunicating duplication.\(^4\) Definitive speculation cannot be made with so few reported cases; however, development of the myenteric plexus could be associated with cystic formation and embryogenesis of the enteric duplication.

In summary, preoperative findings of severely distended cystic formation of the right colon filled with massive feces and associated appendicitis may be considered as a differential diagnosis of right colonic duplication, in both adults and adolescents.

REFERENCES


Fig 2. Intraoperative photograph. The duplication contained both tubular and cystic parts. Arrow, Stump of the appendix. TE, Terminal ileum; Ce, cecum; DT, tubular part of duplication; DC, duplicated colon (cystic part of duplication). Size, 35 × 15 × 6 cm; weight, 2,000 g.