A case of ileocecal duplication cyst protruding into the intestinal lumen enucleated via an anti-mesenteric approach

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Abstract

Ileocecal duplication cysts are generally treated with ileocecal resection and end-to-end anastomosis. However, loss of the ileocecal valve can be a problem in childhood. Cyst enucleation is an option to preserve the ileocecal valve, but is difficult for cysts protruding into the intestinal lumen. We report a case of ileocecal duplication cyst with luminal protrusion successfully enucleated via an anti-mesenteric approach. A 4-year-old boy complaining of abdominal pain was referred to our hospital with a diagnosis of intussusception. Intussusception was reduced with a therapeutic enema, but a tumor was detected in the ascending colon. Ultrasonography showed a cystic tumor adjacent to the ileocecal valve partially sharing the muscle wall with the colon. Surgery was performed with a diagnosis of duplication cyst. The ileocecum was mobilized laparoscopically and exteriorized through the umbilicus. The cyst did not protrude outside the colon, making enucleation difficult. We incised the cecum on the anti-mesenteric side, isolated the cyst, and resected it with preservation of the ileocecal valve. The patient’s postoperative course was uneventful. Nine months after surgery, he remains well, with no sign of recurrence. An anti-mesenteric approach is a good option to preserve the ileocecal valve for ileocecal duplication cysts protruding into the lumen.

Enteric duplication is a rare congenital disorder, occurring in one per 4500 live births [1,2]. Duplication can arise at any level of the gastrointestinal tract from the mouth to the anus, but occurs most commonly in the small intestine [3,4]. Most intestinal duplications lie on the mesenteric side of the intestine, and share a common muscular wall and blood supply with the native bowel. Therefore, the preferred treatment is total cyst excision with resection of the adjacent segment [4–6]. However, when the duplication cyst lies in the ileocecal (IC) region, bowel resection is not ideal, because loss of the IC valve can cause problems in childhood. There have been reports of IC duplication cyst enucleation to preserve the IC valve. However, enucleation of the cyst is difficult when the cyst protrudes into the intestinal lumen. We report a case of IC duplication cyst with luminal protrusion successfully enucleated via an anti-mesenteric approach.

1. Case report

A previously healthy boy, 4 years, 10 months of age, presented at a local clinic complaining of abdominal pain and bloody stool. Abdominal CT scan showed an intussusception. A therapeutic enema was unsuccessful, so the patient was referred to our hospital. On examination, his general condition had improved, with mild right lower quadrant tenderness. Laboratory data showed elevated white blood cell and C-reactive protein levels (25,700 μl−1 and 6.43 mg/dl, respectively). A therapeutic enema was tried again, and...
The intussusception was successfully reduced. However, a tumor was detected in the ascending colon (Fig. 1). Ultrasonography showed a cystic tumor, 3 cm in diameter and partially sharing the muscle wall with the colon. The cyst was slightly hyperechoic, raising suspicion of infection (Fig. 2). Infection was successfully controlled with antibiotics. Surgery was performed with the diagnosis of IC duplication cyst.

Surgery was performed with a laparoscopic procedure via a single umbilical incision. The ileocecum was mobilized laparoscopically and exteriorized through the umbilicus. The cyst did not protrude outside the colon, which made enucleation of the cyst difficult (Fig. 3a). We incised the cecum on the anti-mesenteric side, and isolated the cyst adjacent to the IC valve (Fig. 3b). We circumferentially incised the mucosa at the base of the cyst, and resected the cyst with preservation of the common wall. The mucosal defect was repaired with 5-0 PDS running suture (Fig. 3c, d). Operation time was 146 min; intraoperative bleeding was 47 ml. No transfusion was needed. The patient’s postoperative course was uneventful, and he was discharged on the fifth day after surgery. Nine months after surgery, the patient remained well, with no sign of recurrence.

2. Discussion

Duplication cysts of the alimentary tract can occur at any point in the intestinal tract, but most commonly occur in the small intestine [3]. It is widely accepted that these lesions should be removed completely when diagnosed, because they can contain ectopic tissue such as gastric mucosa, and are associated with malignancy, especially in adults with lesions of the colon [7,8].

The standard surgical procedure for duplication cysts is total resection with the adjacent native intestine. However, for patients with cysts arising near the IC valve, loss of the IC valve can cause problems. The importance of the IC valve is well known. The IC junction is a transition zone regulating intestinal transit from the small bowel into the colon. By slowing the passage of nutrients from the ileum, the IC valve helps to prolong intestinal transit time, thereby increasing the time that nutrients are in contact with the mucosal surface and maximizing absorption of nutrients, fluids, and electrolytes. The IC valve also blocks waste materials from backing up into the small intestine. Removal of the IC valve can lead to bacterial overgrowth, reduced intestinal transit time, and impaired absorption, with symptoms including diarrhea, malnutrition, and electrolyte imbalance [9–11]. Iwanaka et al. reported no significant nutritional disadvantage in children who lost the IC valve with preservation of normal intestinal length [12]. However, the chronic diarrhea caused by loss of the IC valve can be a serious problem [11]. For benign lesions, preservation of the IC region to retain its important function should be considered, especially in children.

Until recently, most IC duplication cases reported in the literature have been treated with IC resection and ileocolic anastomosis. Catalano et al. reported the safety and efficacy of a conservative operative technique to preserve the IC valve in children with IC duplication cyst [13]. Duplication cyst of the intestine usually lies on the mesenteric side of the tract. The cyst is usually approached from the mesenteric side during enucleation. After dissection of the mesenteric sheet above the lesion and mobilization of the cyst from the cecal wall, the cyst is gradually enucleated up to the common wall with the native intestine. Some surgeons incise the shared wall
and close the defect in the native intestine after cyst enucleation [13]. Others preserve the shared wall and resect only the membrane of the duplication cyst.

In our case, the cyst did not protrude outside the intestinal wall, making a mesenteric approach difficult (Fig. 4). In addition, the cyst was just adjacent to the IC valve; resection of the shared wall would make it impossible to close the defect in the native intestine after enucleation. We incised the cecum on the anti-mesenteric side, allowing us to clearly isolate the cyst as a submucosal tumor, and were able to enucleate the cyst with preservation of the shared wall.

There has been a recent increase in the number of reports of laparoscopic or laparoscopy-assisted resection of duplication cysts [5,14]. Cyst enucleation sometimes requires precise procedures to avoid leaving behind cyst membrane. In children, the ileocecal region can often be exteriorized easily through the umbilicus with minimal laparoscopic mobilization, and the cyst can be enucleated with direct visualization. IC duplication cysts are a good indication for laparoscopy-assisted single incisional surgery.

In summary, we experienced a case of IC duplication cyst, successfully enucleated via an anti-mesenteric approach. This

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**Fig. 3.** Intraoperative findings and illustration of cyst resection. a: The cyst protrudes into the lumen, making it difficult to enucleate from the mesenteric side. b: The cyst was isolated as a submucosal tumor adjacent to the IC valve. c: The cyst was enucleated and the mucosal defect repaired. d: The duplication cyst was resected from the mucosal side. After cystectomy, the mucosal defect was sutured.

**Fig. 4.** Illustration of the approach for duplication cyst enucleation. When the cyst protrudes outside the lumen, enucleation is easy. However, when it protrudes into the lumen, enucleation from the mesenteric side is difficult.
approach can be beneficial for IC valve preservation for cysts protruding into the lumen of the intestine.

Conflict of interest
All the authors have no conflict of interest to declare.

References