CASE REPORT

Caecal duplication cyst: a rare cause of neonatal intestinal obstruction

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SUMMARY

We report a rare case of duplication cyst of the caecum responsible for an intestinal obstruction in a 5-day-old newborn. Preoperatively, we suspected the diagnosis of enteric duplication as ultrasonography demonstrated a cystic mass in the right iliac fossa, and laparotomy confirmed an obstructive caecal mass which was resected and an end-to-end anastomosis was performed. The neonate completely recovered with an uneventful follow-up.

BACKGROUND

Intestinal obstruction is the most common surgical emergency in neonates, but it is rarely caused by enteric duplications and is rarely diagnosed preoperatively. Intestinal duplication cysts should be considered in the differential diagnosis in any case of neonatal intestinal obstructions.

CASE PRESENTATION

A full-term 5-day-old male neonate was admitted in the neonatal intensive care unit for a 4-day history of bilious vomiting, abdominal distension and obstipation. The examination found dehydration, a distended tender abdomen and a negative probe test. There was no palpable mass.

INVESTIGATIONS

Abdominal X-ray (figure 1) has shown dilated jejunal and ileal loops with multiple fluid levels. Ultrasonography revealed a cystic lesion around 4×3 cm in size in the right iliac fossa.

Differential diagnosis

- ▶ Alimentary tract duplication.
- ▶ Mesenteric cyst.
- ▶ Meconium pseudocyst.
- ▶ Ovarian compressive cyst.
- ▶ Intestinal atresia.
- ▶ Meconium ileus (cystic fibrosis).

TREATMENT

The patient was optimised for surgery and an exploratory laparotomy was done. Surgical exploration found a dilated ileum 10 cm upstream of an intraluminal caecal mass located on the mesenteric border (figures 2 and 3). We performed an ileocaecal resection with an end-to-end ileocolic anastomosis.

OUTCOME AND FOLLOW-UP

The postoperative recovery and follow-up were uneventful.

Histopathological examination of the resected specimen confirmed the diagnosis of caecal duplication cyst with no ectopic gastric mucosa (figure 4). Now at 7 months follow-up, the patient has normal weight gain without any intestinal complaints.

DISCUSSION

Intestinal duplications are relatively rare congenital anomalies that occur throughout the alimentary tract with a cystic or tubular shape. Approximately 75% of enteric duplications are located within the abdominal cavity, of which only 13% are colonic.¹ The wall's duplication is composed of smooth muscle with gastrointestinal mucosal lining, in about 30% of the cases ectopic gastric mucosa.^{2–4}

Duplication of the caecum is very rare.⁵ Oudshoorn⁵ has reviewed 362 cases of duplication cysts reported in the literature and found only 16

Figure 1 Abdominal X-ray showing multiple fluid levels.

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Figure 2 Preoperative aspect of caecal duplication cyst.

cases of caecal duplications. Presenting symptoms in these cases are as follows: abdominal mass, intussusceptions, gastrointestinal haemorrhage or abdominal pain mimicking appendicitis. It can also cause chronic or acute intestinal obstruction.^{2,4-8}

Duplications of the alimentary tract may present at any age, but 80% present in the first 2 years. In our case, the duplication caused intestinal obstruction in a neonate, which is a rare revelation mode. Although rare, duplication cysts are an important differential diagnosis to consider in children, especially in neonates who present either with intestinal obstruction or with a palpable abdominal mass such as mesenteric, ovarian, pancreatic or choledochal cyst.^{2,8-10}

Preoperative diagnosis of enteric duplication cyst in neonatal intestinal obstruction may be difficult¹¹ and it is often made during laparotomy. The most common imaging modalities to diagnose duplication cysts are ultrasonography and Barium studies, whereas CT and MRI are less often used.⁸ Lower abdominal cystic lesions on ultrasound in case of neonatal intestinal obstruction may discuss the diagnosis of alimentary tract duplication, mesenteric cyst, meconium pseudocyst and ovarian compressive cyst. Our preoperative diagnosis suspected on ultrasound was mesenteric cyst with a differential of alimentary tract duplication, not specifically of caecal origin. Ultrasound demonstrating intra-abdominal cyst should increase the index of suspicion of the diagnosis of caecal duplication cyst. But, nowadays, diagnostic laparoscopy is widely used.¹² In the cases of neonatal intestinal obstruction, although the diagnosis can be made preoperatively by imaging, it should not delay surgery.

The prenatal diagnosis of duplication cysts is increasing, mostly due to the improvement in prenatal screening techniques, routine third trimester screening and improved imaging resolution.^{9,12,13}

Figure 3 The resected specimen showing the duplication cyst within the lumen of the caecum.

Figure 4 Photomicrograph showing histopathology of duplication cysts revealing a double-layered wall with lining of intestinal mucosa.

Learning points

- ▶ Duplication cysts are to be considered in the differential diagnosis in neonates and children who present with acute or subacute intestinal obstruction.
- ▶ Whenever there is a cystic lesion in the right iliac fossa on ultrasonography in a case of a neonatal intestinal obstruction, the first differential diagnosis should be enteric duplication cyst.
- ▶ Complete surgical resection is the treatment of choice as partial excision is associated with high risk of recurrence.
- ▶ Early surgical intervention is important in decreasing morbidity and mortality.

Whenever there is a cystic lesion in the right iliac fossa on ultrasonography in a case of a neonatal intestinal obstruction despite the absence of a palpable abdominal mass, the first differential diagnosis should be 'enteric duplication cyst', whether it is the most common ileal variant, or like in our case, the rarest caecal duplication cyst would be obvious only during laparotomy.

Surgical procedures of small cystic or short tubular duplication involve segmental resection, along with adjacent intestine, with a primary end-to-end anastomosis. If the duplication is a long tubular lesion, it cannot be excised as it will lead to short bowel syndrome. In these cases, selective mucosal excision is an excellent alternative approach.^{2,14,15}

Contributors HA conceived the study, wrote the manuscript and participated in surgery and follow-up. AM operated the patient, supervised the work and reviewed the manuscript. MTC participated in the published work, especially in the writing of the initial draft. YB supervised the work and participated in a critical revision of the manuscript.

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