Rare disease

A common case of gastroenteritis in a child followed by an axial torsion of Meckel diverticulum: a rare and unusual complication

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Summary

A 1-year-old boy presented at our hospital with common gastroenteritis symptoms such as fever, vomiting and diarrhoea. Clinical and laboratory findings were normal. An emergency ultrasound examination was performed and excluded abdominal complications. After 2 days of complete regression of symptoms, the patient began to vomit again, diarrhoea stopped with a sudden worsening of clinical conditions. Laboratory and radiological findings showed signs of an acute abdomen with differential diagnosis between an infectious and an obstructive cause. Owing to the rapid and progressive toxic condition, an emergency laparoscopy was performed. An axial torsion of a swollen and gangrenous Meckel's diverticulum was detected.

BACKGROUND

Meckel's diverticulum has an incidence of 1–2% in the general population. One case out of five develops life-threatening complications that require surgical therapy and about 50% of cases affect children younger than 2 years of age.¹ Axial torsion with gangrene is one of the rarest complications reported till date. Diagnosis before surgery is extremely challenging and the rapidly deteriorating clinical conditions of the patient often require an immediate surgical examination.

CASE PRESENTATION

A 1-year-old boy presented to our paediatric emergency department with 4-day lasting fever followed by vomiting, dehydration and diarrhoea. The clinical examination revealed a mild depletion of body fluids and absence of abdominal pain with no guarding or rebound tenderness. Admission laboratory tests were normal with negative C reactive protein level. Owing to persisting fever associated with thoracic rales, a chest radiograph was performed revealing a mild bronchopneumonia. The abdominal ultrasound showed meteorism and mild intraperitoneal pouring. The patient was admitted to our intensive observation unit and started fluid infusion for gastroenteritis and antibiotic therapy for bronchopneumonia. The following days clinical conditions improved rapidly without further vomiting or diarrhoea, with a decrease in fever and normal abdominal examination. However, on the third day, the clinical conditions worsened suddenly with rebound of high fever. The patient resumed vomiting with occasional emesis and onset of abdominal pain with distension, marked rebound tenderness and diffuse guarding.

INVESTIGATIONS

Laboratory tests were repeated and revealed high white cell count (14 650/mm³ with 69% neutrophils) and C reactive protein level (8.97 mg/dl) with a low level of blood proteins, albumin and electrolytes. The abdominal radiograph showed several air-fluid levels with markedly dilated bowel loops. A second ultrasound examination and an enema with water-soluble contrast ruled out intestinal invagination or other causes of intestinal occlusion. Rectal exploration showed right-sided tenderness with haeme-negative stool. Owing to the sudden worsening of clinical conditions and appearance of more marked abdominal tenderness, the abdominal radiograph was repeated detecting increased bowel distension (figures 1 and 2). This indicated potential small bowel obstruction and thus an explorative laparoscopy was performed, revealing an isolated axially twisted and gangrenous Meckel's diverticulum, 6 cm long with a 3 cm diameter, located 50 cm proximally to the ileocecal valve.

DIFFERENTIAL DIAGNOSIS

Gastroenteritis is one of the most frequent causes of abdominal pain in children and infants. Occasionally, it can be complicated with an abdominal surgical emergency in case of invagination, volvulus and perforation. A case of gastroenteritis complicated by a Meckel's diverticulum is an extremely rare occurrence and is most often mistaken for appendicitis or gastrointestinal haemorrhage or bleeding.² In an acute abdomen phase, the preoperative diagnosis of complicated Meckel's diverticulum is difficult because investigations appear to have no specific value. Nevertheless, abdominal radiography is mandatory in order to reveal signs of bowel obstruction or perforation.

Figure 1 Important bowel distension.

Moreover, ultrasound examination may exclude the presence of intussusception. Sometimes, it is possible to identify an inverted Meckel's diverticulum because it mimics a cystic, tube-like, non-peristaltic structure.³ Such a structure appears as multiple hypoechoic and hyperechoic wall layers with a central hyperechoic area representing serosal fat.⁴

A ^{99m}Tc-pertechnetate scan may be helpful to detect a Meckel's diverticulum only if gastric mucosa is present (15–50% of cases)² but it is not useful in emergency conditions.

Barium enema identifies only 4–22% of the cases but it may be useful to distinguish from other causes of acute abdomen such as intussusception.² Indeed, the diverticulum often has a wide mouth, which allows the barium to empty as fast as it fills; however, the differentiation from an ileal duplication is not possible except at laparotomy.

Figure 2 Air-fluid levels.

Figure 3 The laparoscopic view of the Meckel diverticulum twisted around its axis.

Furthermore, CT-scan images have recently been evaluated in acute abdomen patients with laparoscopic demonstration of Meckel's diverticulum complications: pre-operative diagnosis was performed in 31% of the cases.⁵

TREATMENT

Laparoscopy revealed an isolated Meckel's diverticulum (3 cm×6 cm), axially twisted and filled with secreted fluid, located in the upper right quadrant, 50 cm proximally from the ileocecal valve (figure 3). It was externalised, resected and an end-to-end seromuscular anastomosis was performed to restore small bowel continuity (figures 4 and 5). Contextually, the appendix was also removed. Histological examination confirmed torsion and gangrene of Meckel's diverticulum with leuco-fibrinous perivisceritis (figure 6). No gastric or pancreatic ectopic mucosa was detected.

OUTCOME AND FOLLOW-UP

The second day after surgery the patient showed normal bowel function and after the first 4 days in the intensive care unit, the patient was transferred to the surgery ward

Figure 4 Thanks to the video-assisted procedure, the Meckel diverticulum has been exteriorised through the umbilical incision made to introduce the trocar for the optic.

Figure 5 The end-to-end anastomosis of the small bowel through the 1 cm umbilical incision.

to continue antibiotic therapy and clinical observation. The patient recovered and was discharged 10 days after the surgical operation and no further complications developed during follow-up.

DISCUSSION

Meckel's diverticulum results from incomplete involution of the most proximal portion of vitelline or

omphalomesenteric duct, during week 5 to 7 of fetal development. It is the most common congenital abnormality of the gastrointestinal tract with a prevalence of 1–2% of the population.⁴ It is usually located on the anti-mesenteric border of the ileum within approximately 20–60 cm of the ileocecal valve. Typically, it is short and wide mouthed, on average 2.9 cm long and 1.9 cm wide.⁶ It is a true diverticulum containing all the layers of the intestinal wall. In up to 50% of the cases it also involves ectopic tissue such as gastric mucosa and/or pancreatic tissue. Ectopic tissue is more frequently found in symptomatic cases, most of them in scholar age.² Our patient did not show symptoms related to Meckel's diverticulum during his first year of life most probably because of the absence of ectopic mucosa. It was a large diverticulum (6 cm×3 cm) that represents a rare case itself.

The majority are clinically silent and are incidentally identified during surgery or autopsies. In asymptomatic cases, the male:female ratio is 1.8–2.4:1 while in symptomatic cases male predominance increases from three to five times and 50–60% of the patients are under 2 years of life.^{7–9} The lifetime risk of complications is estimated to be approximately 19%. The complications include haemorrhage (25–50%), inflammation (13–30%) and intestinal obstruction (16%).^{1–8} Occasionally, inversion of Meckel's diverticulum into the lumen of the bowel can cause intussusception, ischaemia and infarction.⁴ When a Meckel's diverticulum is incidentally found at laparoscopy there are no predictive factors for the development of the above described complications. According to the literature and particularly for paediatric patients, the majority of surgeons recommend resection because of the high risk of future complications and the low risk of surgery.⁹

Axial torsion and gangrene of Meckel's diverticulum is the rarest complication that has been reported, particularly in children.^{4–6,10} It was described in only five adults and two children in the past 35 years.^{11–14} The aetiology of axial torsion remains unclear. In some cases, it seems to be caused by the presence of a fibrous cord, a remain of omphalomesenteric arteries, that connect the diverticulum to the umbilicus. However, the majority of Meckel's diverticulum (74%) have a free end.² All adult cases of twisted and gangrenous Meckel's diverticulum are related to fibrous fixed diverticulum, while in children, like in our case, no attachment of the diverticulum to adjacent anatomic structures was identified.

Differently from previously described cases, our patient presented with an initial onset of acute gastroenteritis. We can speculate that the acute infective process stimulated intestinal secretion and peristaltic hypermotility causing twisting of the diverticulum around its narrow base. This generated an ischaemic insult associated with a secondary gangrenous process.

Torsion of Meckel's diverticulum is an emergency condition frequently associated with higher risk of perforation or wider bowel resection. Preoperative diagnosis is challenging since most imaging techniques do not have a strong diagnostic value and only 6–12% of the cases are correctly diagnosed before surgery.¹ Clinical findings are most important for the definition of appropriate surgical timing, and laparoscopy is often the best surgical approach. Indeed, results of surgical management are generally good, with a death rate of 2%.¹

Figure 6 The Meckel's diverticulum was then removed by traditional surgery, resecting the small tract of normal bowel containing it.

Learning points

- ▶ Consider Meckel's diverticulum complications in differential diagnosis of acute abdomen in children with gastroenteritis.
- ▶ In acute abdomen, preoperative diagnosis of Meckel's diverticulum is extremely complex and clinical conditions are the most important elements for definition of the best surgical timing.
- ▶ Laparoscopy is the optimal surgical approach both for correct diagnosis and definitive treatment.

Competing interests None.

Patient consent Obtained.

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