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Congenital Flange Occlusion: About a Case and Literature Review.

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ABSTRACT
Congenital bridle is a rare cause of inclusion in children. It remains difficult to diagnose and is often only confirmed during surgical exploration. We discuss the literature through the case of a 40-day-old girl admitted to the emergency room for an occlusive syndrome and in whom surgery confirmed this rare entity.

1. Introduction

Congenital bridle bowel occlusion (CBO) occurs in a patient with no abdominal, primary or secondary peritonitis and who does not have chronic inflammatory disease of the gastrointestinal tract. The congenital bridle can be derived from embryonic structures such as the vitellin canal, the vitellin artery, the vitellin vein and the ouracus[1]. It may be the consequence of abnormal accotions of the peritoneal foliage during embryogenesis[1]. OIBC is often diagnosed in perioperatively because of its rarity.

2. Observation

Female infant, 40 days old, 3rd of a sibling of 3. Hospitalized for an occlusive syndrome. The onset of symptomatology was 3 days before his hospitalization, by the installation of yellowish vomiting without relation to meals, a material stoppage, a refusal to feed, respiratory grunting, progressive abdominal distension, free hernial openings, everything evolved in a context of apyrexia and alteration of the general state. The clinical examination found a hypotonic infant, mottling of the extremities, and abdominal distension.

After a short conditioning, the thoraco-abdominal radiography was performed, revealed hydroaeric levels wider than high in favour of intestinal occlusion of the hail (Figure 1). The abdominal ultrasound showed a distension of the hailstones and intraperitoneal effusion. The patient had a biological inflammatory syndrome with 12,000 white blood cells/mL and a protein C-reactive protein at 70 mg/L.

The diagnosis of hail occlusion was retained and confirmed during the laparotomy, which revealed 2 tight flanges, with a hail segment of about 30cm in diameter (Figure 2). After section of the flanges (Figure 3), hail appearance improved. In front of the ischemic segment, the option was for a conservative attitude, with a second look in 24 to 48 hours. The second look 48 hours later found the bowel segment to be unsustainable but unperforated, and an anastomosis resection with 30 cm of the middle part of the hail was performed.

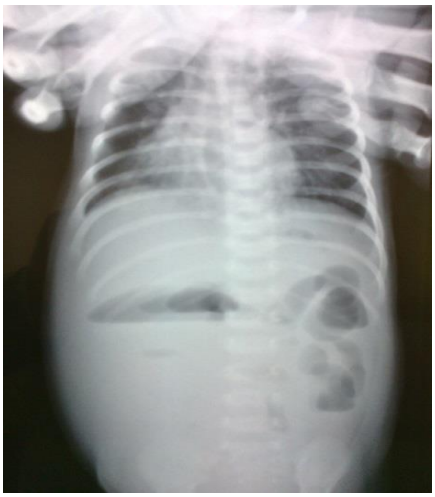

Figure 1. Thoracic-abdominal radiograph showing hydroaeric levels.

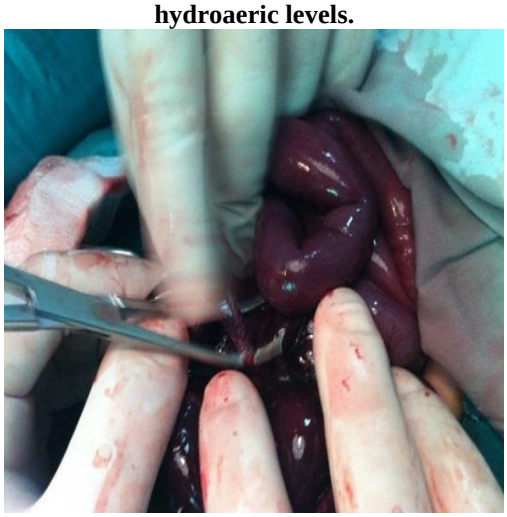

Figure 2. Intraoperative image showing congenital flanges and size disparity.

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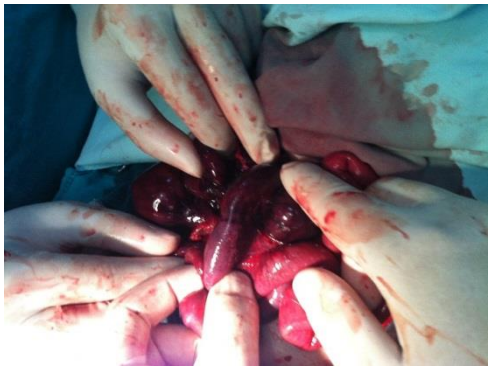

Figure 3. Intraoperative image showing the appearance of hail after section of a flange.

The patient resumed her transit on the 3rd day, feeding was authorized on the 5th day, and she was discharged on the 7th day. Currently, 6 months after surgery the patient is in good health.

3. Discussion

Congenital bridle occlusion of the small intestine is more common in children than in adults[1]. It can result from abnormal adhesion of the peritoneal folds and leaves during embryogenesis, and from the fact that the ventral mesentery flanges remain poorly resorbed[1,2]. The epiploon and the mesentery are the ones most involved in the genesis of the flanges. The small intestine is often more concerned than the colon[1].

The congenital bridle may be responsible for a strangulation of part or all of the small intestine[1-3,4], it may crush the small intestine or cause an occlusive elbow[3,5,7]. Congenital bridle may also be responsible for iterative abdominal pain due to functional pathology[5]. Hail occlusion on the congenital flange is most often abruptly occluded[4]. The most common clinical manifestations are intestinal distension and abdominal pain, the importance of which varies. This pain may be diffuse or localized in the epigastrium or quadrant opposite the strangled handle[5]. Vomiting is first food and then bilious, and the stoppage of materials and gases is inconsistent. Rarely congenital bridle can manifest as chronic abdominal distension and staturoweighteral growth retardation[2]. The time between the onset of the first symptoms and admission varies from a few hours to a few days[5]. The rate of progression depends on the importance of ischemia and intestinal distension.

OIBC produces an acute surgical or sometimes subacute clinical picture, with a clinical picture at admission characterized by diffuse abdominal pain, vomiting, transit cessation, localized or diffuse important defense and diffuse distension of the small intestine[1,5].

The diagnosis of occlusion of the small intestine can be mentioned, but also that of localized or generalized peritonitis. Late diagnosis leads to intestinal ischemia[6].

The unprepared abdomen and the thoraco-abdominal X-ray in the youngest children allow us to appreciate the location and importance of small intestinal distension and its evolution over time[5]. Ultrasound, computed tomography and magnetic resonance imaging show the distended intestine, a strangled handle, and the volvulus of the small intestine, with a sensitivity of 73-95% for computed tomography[2,7,8]. However, due to radiation exposure, computed tomography should not be performed as a first-line procedure in children who consult for an occlusion table[2].

The diagnosis of OIBC is rarely made in preoperatively, whether the clinical picture is acute surgical or subacute and reassuring. When the clinical picture is initially reassuring, there is a risk of letting intestinal necrosis develop. The use of computed tomography, colour echodoppler or magnetic resonance imaging are in these situations of interest for detecting small bowel occlusion or intestinal ischemia[7].

The diagnosis and treatment of congenital flanges is always surgical. Coelioscopic or laparotomy surgery consists of severing the bridle and, if necessary, resecting the necrotic hail loop. Laparotomy is indicated immediately if the small intestine is very distended or if there is intestinal necrosis[9,10]. Extended bowel necrosis can result in a multiviscal failure that is responsible for the patient's death postoperatively.

The evolution is good in case of early diagnosis and early therapeutic management.

4. Conclusion

The occlusion of the small intestine on a congenital flange is a rare condition which poses a real diagnostic problem, especially when the clinical picture is reassuring, with a risk of intestinal necrosis which can be fatal for the patient. The diagnosis is often carried out intraoperatively. The treatment consists of a section of the flange and anastomosis resection in case of necrosis.

Conflict of Interest: NONE

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