CASE REPORTS

PERSISTENT OMPHALOMESENTERIC DUCT: A RARE CAUSE OF ACUTE INTESTINAL VOLVULUS IN AN ADOLESCENT TREATED BY LAPAROSCOPY

PERSISTÊNCIA DO DUCTO ONFALOMESENTÉRICO: UMA CAUSA RARA DE VÓLVULO INTESTINAL NUM ADOLESCENTE TRATADO POR LAPAROSCOPIA

Maria Miguel Gomes¹, Catarina Magalhães Faria¹, Manuela Costa Alves¹, Ana Raquel Silva²,³, IV, Jorge Correia-Pinto²,³, IV, Angélica Osório²,³, IV

ABSTRACT

This report describes the case of a 14-year-old male with a prior history of recurrent abdominal pain and two previous "sub-occlusive" episodes. He presented at the Emergency Room with abdominal pain, emesis, abdominal distension, painful palpation, and signs of peritoneal irritation. Plain upright abdominal radiography suggested a distal small bowel obstruction. Exploratory laparoscopy showed a tubular structure connecting the antimesenteric border of the terminal ileum and the umbilicus, which was behaving as a rotation axis to the small intestine. The volvulus was laparoscopically de-rotated, and the structure was excised. Histological exam confirmed the diagnosis of persistent omphalomesenteric duct.

This case illustrates an intestinal obstruction caused by small intestine volvulus. A high index of suspicion is required to acknowledge this etiology.

Keywords: omphalomesenteric duct; vitelline duct; laparoscopy; small intestinal obstruction; volvulus

RESUMO

É descrito o caso de um adolescente masculino de 14 anos de idade com antecedentes de dor abdominal recorrente e dois episódios “sub-oclusivos” prévios. Recorreu às urgências com dor abdominal e vômitos. Ao exame objetivo, era evidente distensão abdominal e sinais de irritação peritoneal. A radiografia abdominal simples de pé sugeriu a presença de obstrução do íleo distal. Foi realizada uma laparoscopia exploratória, que evidenciou uma estrutura tubular a ligar o bordo antimesentérico do íleo terminal ao umbigo e se comportava como eixo de rotação. Foi efetuada de-rotação do vólvulo e excisão da estrutura. O exame histológico confirmou o diagnóstico de ducto onfalomesentérico persistente.

O presente caso ilustra uma obstrução intestinal causada por vólvulo do íleo distal, sendo necessário um alto índice de suspeição para reconhecer esta etiologia.

Palavras-chave: ducto onfalomesentérico; ducto vitelino; laparoscopia; obstrução ileal; vólvulo

I. Department of Pediatrics, Hospital de Braga. 4710-243 Braga, Portugal. mariamgomes@hotmail.com; catmagalhaesfaria@gmail.com; manuelacostaalves@gmail.com

II. Department of Pediatric Surgery, Hospital de Braga, 4710-243 Braga, Portugal. araquelsilva6@gmail.com; jcp@med.uminho.pt; angelicosorio@gmail.com

III. Life and Health Sciences Research Institute (ICVS). 4710-057 Braga, Portugal. araquelsilva6@gmail.com; jcp@med.uminho.pt; angelicosorio@gmail.com

IV. School of Health Sciences, University of Minho and Portugal Government Associate Laboratory ICVS/ 3B’s. 4710-057 Braga, Portugal. araquelsilva6@gmail.com; jcp@med.uminho.pt; angelicosorio@gmail.com
INTRODUCTION

During embryonal development, the omphalomesenteric duct (OMD) behaves as a primitive connection between the midgut and the yolk sac. The OMD undergoes obliteration and reabsors spontaneously between the fifth and ninth weeks of gestation. Failure of closure or reabsorption occurs in 2% of the population. Different anatomic and morphologic variants of OMD anomalies exist (Figure 1): Meckel's diverticulum, umbilicoileal fistula (patent OMD throughout its length), umbilical sinus (OMD remains open only at its outer portion), umbilical cyst (central cystic dilatation in which the OMD is closed at both ends but patent in the center), fibrous cord (complete obliteration of the duct connecting the ileum to the umbilicus), or umbilical mucosal polyp (persistence of OMD distal end). Meckel's diverticulum is the most common variant, accounting for 65% of cases. Other variants are extremely rare. The affected population can remain asymptomatic until advanced ages. Symptoms are age-dependent and most usually manifest before the age of four (mainly in the neonatal period). Frequency of these entities is similar in both genders, but frequency of symptoms is higher in males. Symptoms include umbilical changes, gastrointestinal obstruction, abdominal pain, and gastrointestinal bleeding, with surgical excision being the treatment of choice.

CASE REPORT

Here is reported the case of a 14-year-old male with prior history of recurrent abdominal pain, constipation, and overweight, who was undergoing a dietary plan, with regular exercise and daily laxative intake. He had two “sub-occlusive” episodes of unknown etiology in the previous three years, which resolved after conservative treatment but required hospitalization, and no previous history of abdominal surgery. No family history of gastrointestinal or other diseases was reported. The boy presented at the Emergency Room with a four-hour history of cramping abdominal pain and emesis (initially bilious emesis but later coffee-ground material), and no further complaints. Intestinal transit was present but decreased. He denied prior trauma and drug or toxic consumption. On examination, abdominal distension, decreased bowel sounds with normal tone, diffuse painful palpation, and mild signs of peritoneal irritation were noted. The remaining physical examination was unremarkable. Plain upright abdominal radiography revealed dilated small bowel loops with air-fluid levels, suggesting a small bowel obstruction (Figure 2). The boy started oral intake restriction and a nasogastric tube drain was placed. Laboratory tests revealed mild leukocytosis (23.9x10^3/µL leukocytes, with 19.9x10^3/µL neutrophils) and negative C-Reactive Protein (<2.90 mg/L). Digestive endoscopy identified no hemorrhagic injury or other changes. An exploratory laparoscopy was performed, which revealed distension of small intestine loops but no macroscopic signs of ischemia. Cecum was normally placed in the lower right quadrant and the colon loops were collapsed. A tubular structure with macroscopic appearance, similar to the small intestine (not just a fibrotic band), was found connecting the antimesenteric border of the terminal ileum to the umbilicus, which was behaving as a rotation axis for the small intestine (Figure 3). The umbilical border was ligated with 2 XL hemolocks, and the volvulus laparoscopically de-rotated. The structure was brought extracelomically and resected with a GIA 55 after ligation of the vitelar artery. Histological examination confirmed the diagnosis of persistent OMD. The post-operative period was uneventful, and the boy was successfully discharged on the fifth postoperative day. He remained asymptomatic over a six-month follow-up period.
Figure 3 - Exploratory laparoscopy. A - Dilated small bowel loops and collapsed colon; B and C - Umbilical fibrous end of the omphalomesenteric duct that was clipped and cut to free the volvulation axis; D - Omphalomesenteric duct basis at the terminal ileum; E - Grasping of the omphalomesenteric duct towards the umbilicus in order to exteriorize it; F - Totally exteriorized persistence of the omphalomesenteric duct and hands-assisted resection with a mechanical suture after identification and ligation of the vitelline artery.

DISCUSSION

Mechanical obstruction is the most frequent surgical problem of the small intestine.\textsuperscript{5,6} Intra-abdominal adhesions related to previous abdominal surgery account for up to 75% of cases.\textsuperscript{5,6} Less prevalent etiologies include hernias, gallstones, neoplasms, intestinal malrotation disorders, or inflammatory pathologies such as tuberculosis or Crohn’s disease.\textsuperscript{5,6} In this case, as there was no history of prior surgery, intra-abdominal post-operative adhesions were rejected as a possible etiology. Presence of two previous episodes of intestinal sub-occlusion and the fact that the patient was followed for recurrent abdominal pain, overweight, and constipation (which was resistant to treatment with symptomatic measures) led to an initial unremarkable investigation comprising laboratory tests, radiographic contrast study, and abdominal ultrasonography.

The initial intestinal obstruction therapy is standard and etiology-independent. Analgesics, oral intake restriction, fluids, electrolyte replacement, and nasogastric drainage are important aspects of supportive care. Broad-spectrum antibiotics may be administered due to risk of bacterial translocation or as prophylaxis for possible resection. This patient followed the initially recommended supportive care and received empirical antibiotics.

Three mechanisms of intestinal obstruction caused by persistent OMD have been reported: intussusception, volvulus, and hernia.\textsuperscript{7-12} The present case illustrates an intestinal obstruction caused by small intestine volvulus. Surgery must be performed early due to risk of strangulation and intestinal perforation and loss. Etiology recognition is difficult without performing exploratory surgery and a high index of suspicion is required.

Clinical presentation is age-dependent and unspecific. Possible symptoms include irritability, umbilical discharge, umbilical hernia, emesis, abdominal pain, gastrointestinal bleeding, and absence of intestinal transit. Initial laboratory tests may be normal or show elevated inflammatory parameters. Abdominal radiography and ultrasonography are unspecific. Abdominal computed tomography may occasionally evidence the presence of a fibrotic band connecting the umbilicus to the small intestine. In this case, an abdominal computed tomography was not performed because the patient presented with an acute abdomen and such exam would delay treatment. Diagnosis was only possible through surgical exploration. Surgical excision of the vitelline duct anomaly is considered therapeutic.

Intestinal obstruction by persistent OMD is extremely rare.\textsuperscript{7-9,13-15} Early surgical resection by laparotomy is performed in symptomatic patients in most cases described in the literature.

Laparoscopy is associated with several technical difficulties, mainly due to bowel distension. Presence of a structure connected to the umbilicus may pose some problems when using the umbilicus as a working channel for laparoscopic instruments. Nevertheless, we consider that laparoscopy should be the preferred choice for approaching these patients due to the advantages of a minimally invasive surgery, including reduced pain and infectious complications, early recovery, and better cosmetic results.
REFERENCES


CORRESPONDENCE TO

Maria Miguel Gomes
Department of Pediatrics,
Hospital de Braga
Sete Fontes 4710-243
São Victor, Braga
Email: mariamgomes@hotmail.com

Received for publication: 22.10.2017
Accepted in revised form: 19.12.2018