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An association between transmesenteric internal hernia and abdominal cocoon syndrome: a case report

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ABSTRACT:

INTRODUCTION: Transmesenteric internal hernia is defined as the herniation of the small intestine from a mesenteric defect in the abdominal cavity, and abdominal cocoon syndrome is the partial or entire encapsulation of the small bowel like the shape of an accordion, by a fibrocollagenous membrane.

PRESENTATION OF CASE: A 32-year old male patient applied with complaints of abdominal pain, nausea, and vomiting bile. Signs visualized in the abdominal computer tomography were as follows: gatto formation of the small intestinal loops and suspected of an internal hernia. In the operation, a membrane was detected encapsulating the entire intestine resembling a tube, making the intestines to appear like an accordion and an opening was present in the small intestinal mesentery. The intestine was separated from the defect, and placed in its normal anatomical position. The defect in the mesentery was closed and the encapsulating membrane was removed from small intestine. Intestinal resection was not required.

DISCUSSION: Internal hernias comprise less than 1% of all intestinal obstructions, and are formed by the herniation of the intestine and mesentery into the opening of the visceral peritoneum or into the recessus. Abdominal cocoon syndrome is a disorder characterized by the partial or total encapsulation of the small intestine by a thick and fibrotic membrane. Preoperative diagnosis is very difficult and is generally diagnosed during laparotomy exploration.

CONCLUSION: The association of internal herniation and abdominal cocoon syndrome is an extremely rare cause of mechanical intestinal obstruction. If not promptly diagnosed and treated, can lead to serious complications.

Keywords: Abdominal cocoon syndrome, internal hernia, ileus, acute abdomen, case report

Introduction

Internal herniations are defined as the protrusion of small intestinal segments into the anatomical or non-anatomical spaces within the peritoneum, and they include less than 1% of cases with intestinal obstruction (1,2). Intestinal obstruction associated with internal herniation may exist in any age; however internal hernias due to congenital transmesenteric defects are mostly reported in pediatric age groups and frequently concomitant with intra-abdominal anomalies like intestinal atresia. In adults the defect is mostly acquired as a result of blunt abdominal trauma or previous intestinal and mesentery surgeries (3). In the absence

of an anamnesis, revealing a previous abdominal surgery, a preoperative diagnosis of these herniations is difficult and therefore rapidly progress to a life-threatening intestinal ischemia. Due to delays in diagnosis and treatment mortality rates can rise up to 20% (4).

Abdominal cocoon syndrome is a rare condition, which is the partial or complete encapsulation of the intestines by a fibrocollagenous membrane resembling an accordion, existing as an acute or subacute intestinal obstruction. Mostly observed in young adolescent girls, it was first defined in 1868 and exists with non-specific signs, generally diagnosed during surgery (5,6). Causes of fibrosis have been claimed in its physiopathology and include the retrograde menstruation, previous abdominal surgery, use of beta-blockers, peritoneal dialysis, cirrhosis, Lee Veen shunt and idiopathic cases exist congenitally in which no cause leading to cocoon formation can be determined. Both transmesenteric internal herniation and abdominal cocoon syndrome causes of intestinal obstruction that rarely exist on their own.

We present here an extremely rare case of an association of transmesenteric internal herniation and abdominal cocoon syndrome, which we operated on due to signs of acute abdomen and mechanical intestinal obstruction, and was diagnosed during the operation.

Case report

A 32-year old male patient applied with complaints of abdominal pain, nausea, and vomiting bile, which had started the previous day. His medical history revealed absence of a known chronic disease or drug use, undescended testicle surgery at 8 years old, and a defecation habituation of once every two days. His family history showed no specific property. His physical examination body temperature 37.9°C, pulse 100/min, blood pressure 120/70 mmHg, and respiratory rate 16/min. A slight distension, and a generalized tenderness that was more marked in the epigastric area were detected in the abdominal examination; abdominal defense was absent and rebound phenomenon was found to be positive. Direct abdominal x-ray taken in the upright position revealed several air-fluid levels. Abdominal ultrasonography evaluation revealed no pathological sign, except a slight dilation of the intestinal loops. Laboratory analysis revealed the following: leukocyte 6570/mm³, Hb 14.2 gr/dl, Plt 246000/mm³, CRP 8.9 mg/L. In abdominal CT minimal fluid in the perisplenic area, gatto formation of the small intestinal loops in the abdominal midline and left lateral and an image in the right side suspected of an internal hernia (**Figure 1 and 2**).

The patient underwent surgery for the mechanical intestinal obstruction. The small intestine was dilated in appearance and marked abdominal adhesions were observed. A membrane was detected encapsulating the entire intestine resembling a tube, making the intestines to appear like an accordion (**Figure 3**). In the right, an opening was present in the small intestinal mesentery; the rest of the small intestine was observed to herniate from this opening, together with the encapsulating membrane (**Figure 4**). The intestine was separated from the defect, and placed in its normal anatomical position. The defect in the mesentery was closed. During the first exploration the length of the small intestine was observed about one meter; this length was discovered to be 3.5-4 meters following the removal of the encapsulating membrane. Intestinal resection was not required.

Histopathological examination of the membrane revealed fibrous connective tissue with chronic inflammation, congestion, and edema, showing scleral hyaline changes. Bowel movements began on the post-operative fifth day. The patient was discharged from the hospital with recovery, on the post-operative seventh day. In his follow-ups after hospitalization, no abnormalities existed in the gas or feces discharge; however, he experienced loss of appetite, nausea, vomiting, indigestion, and about 8 kg of weight loss. His complaints lasted for nearly two months, showing a gradual decrease. In the period following these two months, he felt relieved, and regained his previous weight.

Discussion

Internal hernias comprise less than 1% of all intestinal obstructions, and are formed by the herniation of the intestine and mesentery into the opening of the visceral peritoneum or into the recessus (2). Internal herniations are so named due to the openings they pass through, or the position of the herniated organ. According to these considerations, they exist in different types, as paraduodenal, foramen of Winslow, pericecal, intersigmoid, transmesenteric, transomental, and retroanastomotic. The type most frequently seen is the paraduodenal hernia, which forms 53% of all internal herniations (7). In some series, the transmesenteric type is more frequently observed in children (4). When replaced and gathered segments of the small intestine, and rotational movements of the mesenteric vascular structures supplying these intestinal segments are visualized in an abdominal CT, internal herniation has to be suspected. The time duration for the progress of an intestinal obstruction to the intestinal ischemia may be short. Therefore, losing time through conservative treatment may be life threatening in these cases. The clinical status of the patient has to be the primary guide when deciding on surgery. Morbidity and mortality can be reduced by a last minute intervention (4).

Abdominal cocoon syndrome is a disorder characterized by the partial or total encapsulation of the small intestine by a thick and fibrotic membrane (8), incidences of which are reported to occur between 0.4% and 5.5% (9). Idiopathic abdominal cocoon occurs congenitally, in which no cause leading to cocoon formation can be detected (8). Secondary abdominal cocoon formation is determined more frequently, as a result of abdominal surgery, the use of beta-blockers (practolol), peritonitis related with peritoneal dialysis, Lee Veen shunt, cirrhosis and irregular menstruation. Its etiology includes various acquired factors; however the common fact for all is the existence of a condition that leads to an intraabdominal reaction (8,10). Preoperative diagnosis is very difficult and is generally diagnosed during laparotomy exploration (7). Laboratory findings do not reveal any sign of it being specific to the abdominal cocoon(11). Routinely taken direct abdominal graphies in the upright position do not indicate a specific sign, except the gas-fluid levels(11). In CT, the gathering of small intestinal segments in a single area, and a membrane with soft tissue density surrounding them, are the classic signs. Other CT signs include the collection of ascites or locular fluid, peritoneal thickening and contrast trapping, peritoneal calcifications, and the thickening of the intestinal wall (11).

As a conclusion, the association of internal herniation and abdominal cocoon syndrome is an extremely rare cause of mechanical intestinal obstruction. Its treatment includes the excision

of the cocoon by careful dissection, the reduction of the hernia, the resection/anastomosis of the devitalized intestine, and repair of the mesenteric defect. Time duration for regaining the small intestinal functions is longer in these patients compared with other cases with ileus, and these functions become regular over a longer time. If not promptly diagnosed and treated, can lead to serious complications such as intestinal ischemia, perforation, sepsis and death. The most important goal of management is early identification of the disease which provides a better prognosis.

This case report is compliant with the SCARE Guidelines and criteria (12).

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Figure Legends:

Figure 1. A section of the axial computed tomography; thickened membrane is visualized, surrounding the conglomerated intestinal loops.

Figure 2. Small intestinal loops create gatto formation in the abdominal midline and left lateral; in the right, an image is visualized which is suspected of an internal hernia.

Figure 3. Intraoperative appearance of membrane surrounding the small intestine in the abdominal cocoon syndrome.

Figure 4. Appearance of the transmesenteric defect, and herniated small intestine from this defect.

Fig.1



Fig.2



Fig.3

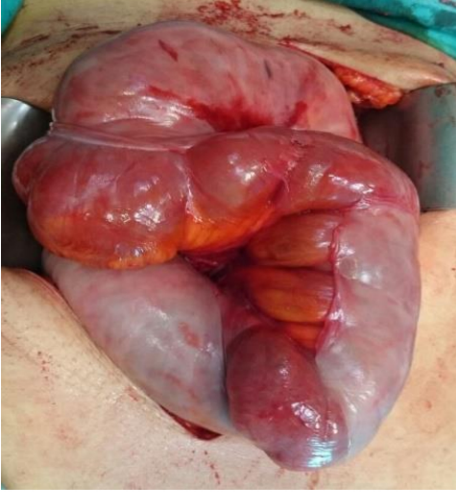


Fig.4

