A Rare Case of Abdominal Cocoon Presenting as Umbilical Hernia

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Abdominal cocoon (AC) is a rare condition, that leading to acute or chronic small bowel obstruction, characterized by a total or partial encapsulation of the small bowel by a fibrous membrane or sac-like cocoon, sometimes colon, uterus or accessories are encased in. The disease is characterized as either primary or secondary to other causes. The main reported clinical manifestations of AC are acute/subacute complete/partial intestinal obstruction and abdominal mass.[1] Here, we report a rare case of AC presenting as umbilical hernia, and to our knowledge, it is only a few cases of this entity have been reported previously. Even though preoperative diagnosis of AC is difficult and normally laparotomy is the main solution, in our case, we chose the method which combines laparoscopy with open surgery, it solved patient problem quickly and efficiently, and that further confirmed laparoscopic surgery has great significance in diagnosis and treatment of AC.

A 40-year-old male presented to our department complaining periumbilical mass and abdominal pain. He had a clinical history of gradually increased mass in the umbilicus region for 2 years without pain, but recently he felt colicky abdominal pain over the past 2 months, which occurred mostly around the umbilicus. The symptom came along with abdominal distension, vomiting, belching, sour regurgitation, constipation, and body gas reduction. The patient had inguinal hernia repair operation at 2-year-old, and he had no other special medical history, such as peritonitis, tuberculosis, or chronical drug intake. His general physical examination revealed a chronic face and mild emaciation. Local abdomen examination revealed a distended abdomen, mild tenderness, and rigidity in the whole abdomen while bowel sound was normal. A tender round-shapes lump was inspected and palpated in the supraumbilical region which was approximately 4 cm in diameter. The rectal examination was normal. Routine laboratory studies demonstrated no abnormalities. His enteroscope result came up normal, X-ray of the abdomen showed intestinal obstruction [Figure 1]. B-ultrasonic wave revealed a hypoechoic periumbilical abdominal mass referred to umbilical hernia. Computed tomography (CT) scan demonstrated umbilical hernia and an incomplete low small bowel obstruction without clear cause. Moreover, part of the bowel wall became thickening: A possible inflammatory lesions and pelvic effusion were involved as shown in Figure 2. He was suspected with an umbilical hernia and partial intestinal obstruction. Laparoscopic exploration was performed after a complete preoperative preparation. Using laparoscope, we found that small bowel was covered with a dense whitish fibrous sheath, which gave the appearance of a cocoon. Hence, the clear diagnosis was AC. We then moved to laparotomy, that omentum majus absent had been detected, the entire small intestine was wrapped by a tubular white capsule to about one meter localization [Figure 3], small mesentery and colon have the similar lesions, to a lesser degree. The coated surface was smooth and easy to separate from the intestinal wall, there was film adhesion seen in the apart small intestine [Figure 4]. We gradually stripped the coated membrane, released the small intestine, to make sure the small intestine regained its normal anatomy status and location, and we did appendicectomy at the same time that appendix was obvious fibrosis, umbilical hernia repair was carried out in the end. The excision's appearance looked like milky white, cocoon-like fibrillar connective tissue [Figure 5]. Before closing the abdominal wall, we injected medical sodium hyaluronate gel to prevent postoperative recurrence of adhesions in the abdominal cavity. The patient had a comprehensive symptomatic treatment after the operation. The peeled-off membrane was studied histopathologically and revealed dysplasia of the dense fiber membrane, a chronic nonspecific inflammatory

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reaction and chronic appendicitis [Figure 6]. Eleven days after the operation, the patient recovered well and discharged from the hospital.

Abdominal cocoon was first observed by Owtschinnikow in 1907, who called it as peritonitis chronica fibrosa incapsulata at that time, first described and named by Singapore researchers Foo *et al*. in 1978. The etiology and pathogenesis are still unknown, it is considered as a form of chronic irritation and inflammation. There are two types of AC – primary or idiopathic, and secondary.

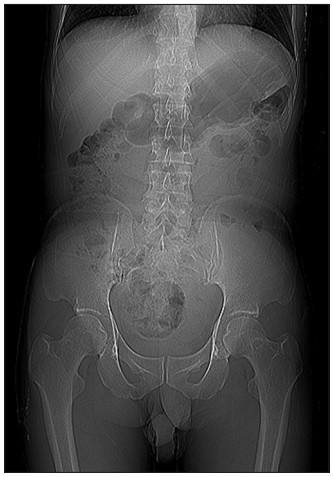

Figure 1: X-ray plain abdominal imaging revealed intestinal obstruction.

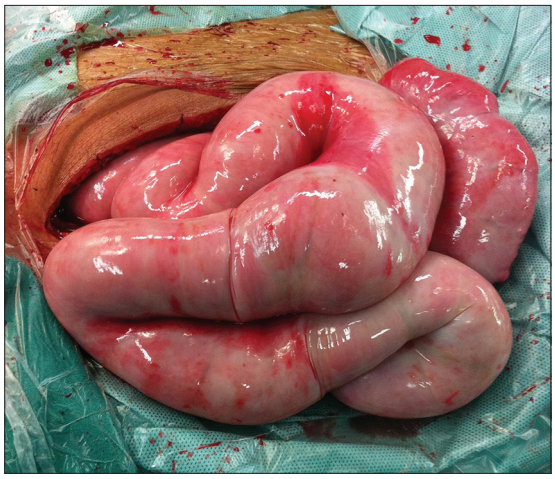

Figure 3: Intraoperative photographs show the encapsulated small bowel segments with a dense fibrous layer.

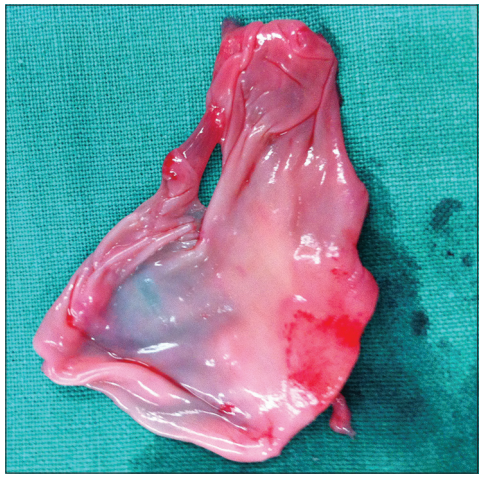

Figure 5: The excision's appearance.

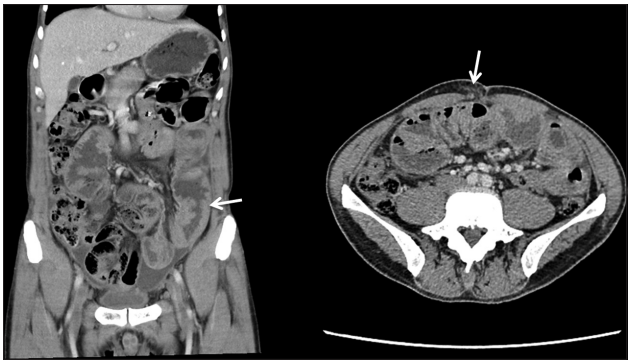

Figure 2: Computed tomography shows umbilical hernia and an incomplete low small bowel obstruction, part of the bowel wall became thickening, and pelvic effusion.

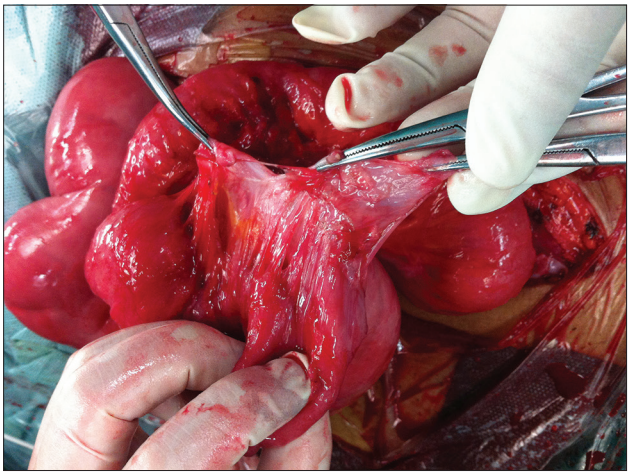

Figure 4: Intraoperative photographs show the film adhesion in the apart small intestine.

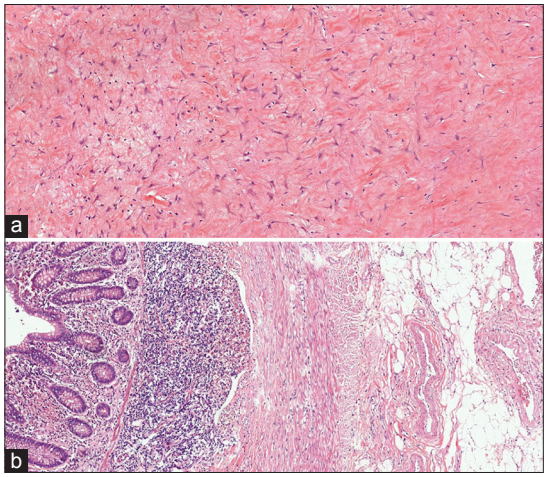

Figure 6: (a) Histological examination showed the dysplasia of the dense fiber membrane, a chronic nonspecific inflammatory reaction and (b) showed chronic appendicitis.

The primary form of the disease is more common and has been described mostly in young adolescent females from the tropical and subtropical countries, but also males, premenopausal females, and children can be affected in this condition. However, the exact stimulus for the inflammatory reaction is unknown. The secondary form has been reported in associated with a history of previous abdominal surgery or peritonitis, prolonged use of practolol, chronic ambulatory peritoneal dialysis, ventriculoperitoneal shunts, intraperitoneal instillation of drugs, sarcoidosis, SLE, liver cirrhosis, hysteromyomas, ovarian endometriotic cyst or tumor, and tuberculous etiology.[2,3] For this patient, although he underwent inguinal hernia surgery at a young age that was not enough to induce inflammation after so many years making peritoneal fibrin extensive sheep and forming capsular parcel causing AC, but with omentum majus absent, the cause of this case considered as congenital factors. Our patient hospitalized by umbilical hernia with small intestinal obstruction, with suspicion we finally diagnosed him with AC. It is really rare that AC presented as umbilical hernia. We inferred that this patient is a primary AC patient; it follows that congenital AC, which may not be a single disease, but could cause other organs hypogenesis or diseases, such as umbilical hernia, etc. The signs and symptoms are not just presented as intestinal obstruction which needs to be investigated seriously.

Although there are no specific clinical symptoms, and lack of specific preoperative examination demonstration, preoperative diagnosis of AC is still possible. It needs to rely on the current imaging examination which is very useful for diagnosis and differential diagnosis, such as B-ultrasound wave, CT, gastrointestinal imaging. CT is very helpful for AC, in our case preoperative CT showed bowel wall thickening and small-bowel obstruction, this can be highly suspected as AC. If CT shows thick enhancing membrane surrounded the bowel, forming a saclike structure or a cocoon, the diagnosis of AC can be confirmed.[4]

Surgery is the most fundamental way to solve this problem, careful dissection, and excision of the thick/thin sac-like membrane to release the small intestine leads to complete recovery. Some cases have been reported using laparoscopic examination to handle it,[5] from our case, we can see that laparoscopy can easily check out of AC, it further illustrates laparoscopic surgery is really a worthwhile method for the diagnosis of AC. Thus, total laparoscopic surgery in the treatment of AC, may prolong the postoperative intestinal recovery time, because of the increasing operation time and a large number of pneumoperitoneum stimulation. In our case, we combined laparoscopy with open surgery to cure the patient, rapid diagnosis at first, and then quickly and efficiently remove the patient's pain; this combination method is worth promoting. We performed appendectomy in our case; it can avoid the patient to develop acute appendicitis in the future to increase the chance of re-operation. In our opinion, an incidental appendectomy is recommended with or without appendix lesions. We injected some prevent or reduce intestinal adhesion material to the abdominal cavity before abdominal closing, hoping that this material can work out to prevent membrane reproduce but does it really work? We cannot make a precise answer since AC has a low incidence, and we do not have a good amount of clinical samples to do statistical analysis and comparison to confirm the result. It is a significant research, which can make contributions to AC patient in future. Our patient received a comprehensive treatment postoperation, he reviews regularly since hospital discharge for more than 1 year, and no symptoms recur.

Here are the experiences of this case: (1) Fully imaging examinations should be done for unknown reason intestinal obstruction patients, like CT, abdominal plain film, barium meal, etc. If coated disease discovered, there is AC, and it should be highly suspected if bowel becomes thicker; (2) Laparoscopy could be a better method for discovery. We should resect thick membrane to remove the obstruction, along with appendicectomy; (3) In postoperative period, use various means to stop adhesions happening again.

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