Case Report

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Volvulus of the jejunum on cystic lymphangioma: About a clinical case

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ABSTRACT

Intestinal volvulus on mesenteric cysticum lymphangioma (CL) is rare in children. The clinical picture is not very suggestive. We report a case of intestinal volvulus on CL in a 7-year-old girl after an abdominal trauma. Resection and anastomosis were made. The confirmation diagnosis was done by anatomopathological examination. Early diagnosis of intra-abdominal CL will allow avoiding complication.

Key words: Cysticum lymphangioma, intestinal volvulus, surgery, trauma

INTRODUCTION

Cysticum lymphangioma (CL) is conjunctival malformative vascular tumours corresponding to a detention of lymphatic tissue due to an abnormal embryonic development of the lymphatic system.[1] Intra-abdominal forms are rare and are preferentially situated in the mesentery.[2] Its clinical signs are related to the tumoural volume or a mechanical, infectious or haemorrhagic complication. Inaugural intestinal obstruction is rare, exceptionally by direct compression, more probably by twisting, leading to a small intestinal volvulus.[3] We report the case of abdominal localisation of a child of 7 years with inaugural occlusive manifestation after blind abdominal trauma.

CASE REPORT

Miss XY, 7 years old, has been sent by Mother-Child Hospital on 12th October 2014 for diffuse abdominal pain. This symptomatology appeared further to an accidental fall on class' bench 2 days previously. Associated signs were stop of material and gas, bilious postprandial vomiting and anorexia, with hyper sweating. Upon physical examination, the child presented change in the general state, dehydration folds, with a weight of 20 kg, a heart rate of 120 beats/min, a respiratory frequency of 30 cycles/min and a temperature of 37.8°C. Belly was slightly distended, sensitive and no tender without tangible mass. A plain abdominal X-ray had realised before her evacuation, allowed highlighting water and gas levels higher than wide making evoke a small intestine occlusion. Rate of haemoglobin was 11.8 g/dL, haematocrit 35.5% and Group A positive.

Diagnosis of an intestinal obstruction was made. Resuscitation with normal saline 0.9% was realised 2 h before surgery. After clinical improvement, she was taken to operating room for the surgical operation. Under general anaesthesia, a median laparotomy was realised. Upon exploration, there was volvulus of the jejunum on a multicompartmentalised cyst developed in the intestinal wall [Figure 1], with necrosis of twisted intestine by compression [Figure 2]. Resection and anastomosis were made. At macroscopic examination, specimen was in the form of multiple compartmentalised, translucent cysts containing blood liquid. Histologically, these cavities were optically empty, limited by fine collagenic wall containing slender smooth muscular fibres and lined on endoluminal

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hillside by a laminated close cover of devoid flattened cells of atypies cyto-nuclear power [Figure 3]. Diagnosis of benign mesenteric multicystic lymphangioma was confirmed. Operating suites were simple, the patient got back her transit the third postoperative day and food was authorised. She went back home the 6th day of surgery.

DISCUSSION

Lymphangioma is a rare benign lesion in children. It represents 7% of the intra-abdominal cystic formations with preferential ileal localisation.[3] Its affects four times as many boys as girls.

Arrested development of lymphatico-venous connections during the embryogenesis would be the cause of its emerging. The absence of drainage of the primitive lymphatic bags would be responsible for the formation of a cystic lesion containing some lymph. The abnormal obstinacy of the retroperitoneal bag would so be at the origin of retroperitoneal mesenteric or meso-colonic localisations and after its attraction forward during the development of Mesos. This congenital theory is strengthened by observations of CL detected in the prenatal period. However, the diagnosis can be made at any age.[4]

The clinical evolution seems different according to the age of the patient, the volume of the cyst and/or its complications. Intestinal obstruction related to the mesenteric cysts can be induced by compression or traction by the mass.[5] There are two theories explaining the relevance between small bowel volvulus and lymphangioma. The first is that the flaccid and mobile characteristics of a mesenteric lymphangioma cause it to rotate, which induces small bowel volvulus. The second is that longstanding or intermittent volvulus causes lymphatic obstruction, which forms lymphatic cysts as a result. The latter case generally forms a unilocular cystic mass without internal septa.[6] In our case, a multilocular mesenteric cyst was the primary pathology that secondarily caused volvulus of the small bowel after trauma. A plain abdominal X-ray realised was not able to show the cystic mass.

The diagnosis of certainty is made by the anatomopathological study of the operating specimen which objectifies multiple cystic cavities optically empty limited by a fine collagenic wall containing slender smooth muscular fibres and lined on endoluminal hillside by an unistratified cover. The immunohistochimical study confirms the endothelial nature of this cover as well as the constant presence of smooth muscular fibres. The differential diagnosis can be made with a multicystic mesotheliom, a benign, often widened, diffuse and recurrent but also with a lymphangiomatosis, a lymphatic

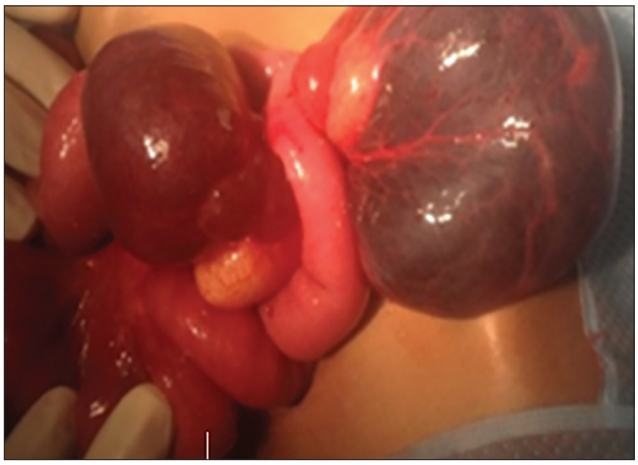

Figure 1: Volvulus of the jejunum on a multicompartmentalised cyst

Figure 2: Necrosis of twisted intestine

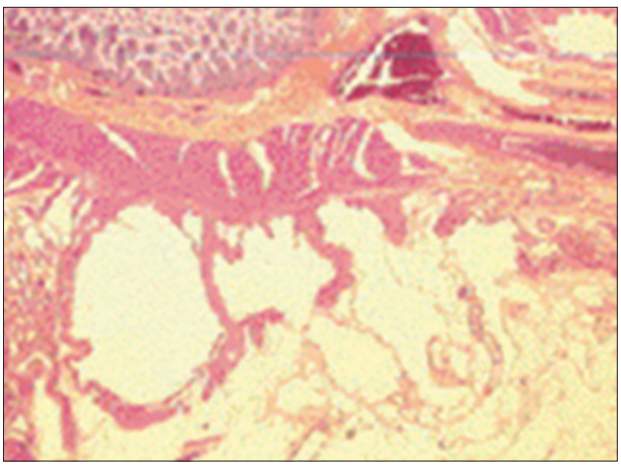

Figure 3: Fine collagenic wall containing slender smooth muscular fibre

compensation of angiomatoses, achieving in a diffuse and concomitant way, viscera, soft tissue and osseous structures.[7]

Ideal treatment of CL is surgery with complete excision because there is a high risk of evolution of the lesion (increase in volume, infection, bleeding and volvulus). This resection is often easy and realised by coelioscopy when it is pediculate, without proximity with the bowel.[8] Because of anatomical relationships with a very close vascularisation or when it is too voluminous, the resection of an intestinal segment can be imperative, as shown in our observation.

Nevertheless, there is a rate of recurrence of 40% after incomplete resection and after 17% after a complete macroscopic manner of excision.[9]

CONCLUSIONS

CL can lead to small intestine volvulus and grave prognosis. It is thus essential to make diagnosis of this mass to avoid an abdominal complication.

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Conflicts of interest
There are no conflicts of interest.

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