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Erroneous Diagnosis of an Inguinal Cystic Lymphangioma as being an Inguinal Hernia: A Case Report

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Abstract **Case Report**

Cystic lymphangioma is a rare benign tumor of the lymphatic system with highly pleomorphic clinical features that mainly affects children. Cystic lymphomas can develop in different anatomical sites, resulting in an equivocal clinical presentation, rarely manifesting in the inguinal canal. Diagnosis is mainly based on abdominal ultrasound, supported by computed tomography. However, only pathology can confirm the diagnosis. Complete surgical resection is the treatment of choice. Cystic lymphangioma has a good prognosis overall, with an exceptionally low recurrence rate.

Keyword: Inguinal, rare, diagnosis, cystic lymphangioma, hernia, tumor.

INTRODUCTION

Cystic lymphangioma is a rare benign tumor of the lymphatic system [1, 2]. It is rare in adults but usually seen in children and exceptionally in adults [3]. Scrotal and inguinal localization remains rare [4]. The clinical presentation is polymorphic friction. Craniofacial, cervical or axillary localization is the most common whereas intra-abdominal forms are rare. They preferentially affect the mesentery, omentum but also the liver, spleen, pancreas, kidney, adrenal gland, colon and duodenum [5]. Localization in the abdominal cavity is in the range of two to 10% [6]. The diagnosis is evoked by imaging but requires histological confirmation. Surgery by total removal remains the most effective alternative with the least recurrence [6].

CLINICAL CASE

Patient aged 20 without any particular history, presenting for a pain of the right iliac fossa evolving for more than 24 hours before the admission with stop of matter and gas; within a febrile context quantified in 38, 5°C. Clinically, a painful mass in the right inguinal region, irreducible and non-expansive when coughing. The rest of the physical examination was unremarkable. The white blood cell count was 22x109/L, the CRP was 180mg/L and the hemoglobin level was 12 g/dL. The rest of the biological examination was without particularity. An abdominal ultrasound was therefore performed, showing a subcutaneous cystic formation of roughly oval shape, with regular contour, well limited, without septum, not lighting up on color Doppler. Also , the abdominal CT scan showed a cystic formation of the left inguinal canal measuring 18x26x49 mm (APxTxCC) with no continuity with the digestive or gynecological structures, associated with a minimal pelvic effusion and no visible endocytic buds (Figures 1 and 2). In view of the painful nature of the swelling and the inflammatory signs opposite, the patient was taken to the operating room and the surgical exploration objectified the presence of a cystic formation on the inguinal region adherent to the neighboring structures with liquid content and an inflammatory reaction around this formation; a meticulous dissection was performed with resection of the cystic mass. The postoperative course was simple, and the patient was declared discharged on day 3 of the postoperative period. The result of the anatomopathological examination objectified a fibrous wall that could fit into the framework of a cystic lymphangioma. The patient was seen in consultation at 4 months hindsight with a normal inguinal region control ultrasound.

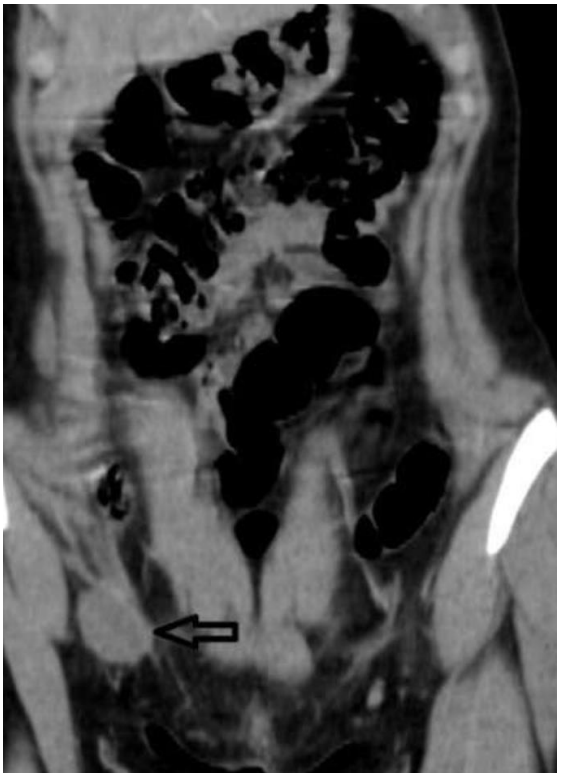

Figure 1: CT appearance of the cystic formation of the right inguinal canal (arrowed) measuring 49 mm in long axis with no continuitý with digestive or gynecological structures

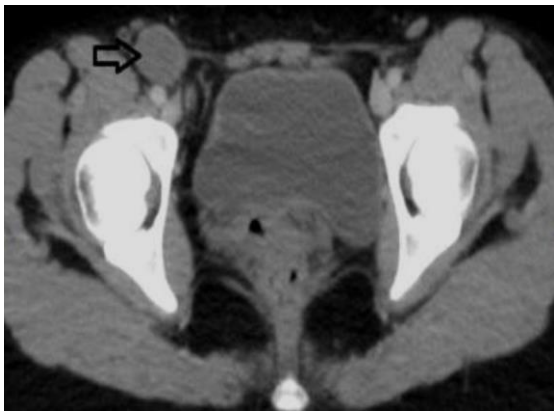

Figure 2: CT appearance of the cystic formation of the right inguinal canal (arrowed) measuring 49 mm in long axis with no continuitý with digestive or gynecological structures.

DISCUSSION

Cystic lymphangiomas are benign congenital tumors of unknown etiology. The majority of lymphangiomas (90%) develop during the first two years of life and 50% are present at birth [7]. Development in adults is exceptional. Males and females are reported to be equally affected in adulthood, whereas in children, the sex ratio is either similar or slightly predominant in boys [6]. Inguinal and scrotal lymphangiomas are rare. Epididymal lymphangiomas are extremely rare with only 6 such cases reported in the literature [8, 9] even rarer are those of the inguinal region. A cystic lymphangioma usually presents as a mass that progressively enlarges with time [7]. Sometimes it presents with an acute onset of pain with rapid and sudden enlargement [7]. This occurs after a hemorrhage, an inflammation, which was the case in our patient, or a disruption of the balance between lymphatic production and drainage [10]. Ultrasound usually reveals a multi-cystic, non-vascularized mass. Some cysts may contain fine echogenic material corresponding to blood [3]. CT is an excellent initial diagnostic tool with adults [11, 12]. It usually shows a homogeneous, hypodense tumor that does not take contrast, as well as its thin septa, and it allows to study the density of the tumor. The CT scan also allows to evaluate the dynamic interaction of the tumor with the surrounding organs. In our case, it allowed to specify the relationship of the cystic formation and its origin especially in the context of emergencies. Magnetic resonance imaging (MRI), as a second line, was not performed on our patient given to the context of the emergency, allows for a better clarification of the nature of the cysts contents and appreciates very well the perivascular extension of the lesion. The cystic lymphangioma is fluidic in signal: hyposignal in T1 and hypersignal in T2. The septa are hyposignal in T1 and T2. Gadolinium injection shows little or no parietal and septal enhancement [6]. The definitive proof of the diagnosis of cystic lymphangioma is provided by anatomopathological examination [11, 12]. The treatment of choice is surgical, consisting of a complete removal of the lesion [3]. The excision must be as complete as possible to avoid recurrence [11]. The recurrence rate is 40% after incomplete resection and 17% after macroscopically complete resection, all locations combined [12].

CONCLUSION

The cystic lymphangioma is a rare benign tumor, which usually occurs at the age of childhood and rarely being adult. The inguinal localization is rare and can take the form of a strangulated inguinal hernia in case of complications. The diagnosis is made in front of the ultrasound and the scanner allows to study well the reports of the lesion. The anatomopathological gives the diagnosis with certainty and the treatment of choice remains the surgery with complete exeresis of the tumor to avoid the recurrence.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

CONTRIBUTIONS OF THE AUTHORS

F.S. Ondongo: author, M. Boui: co-author. All authors have read and approved the final version of the manuscript.

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