Infected retroperitoneal cystic lymphangioma: a case report

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Section: Abdominal imaging
Case Type: Clinical Cases
Patient: 62 years, male

Clinical History:
In this report we describe a case of an infected retroperitoneal cystic lymphangioma, a rare disease in adults. The physiopathology features, diagnostic approach and treatment are described.

Imaging Findings:
A 62 year old man came to our emergency department with a 10 day history of abdominal right flank pain, general malaise and anorexia with a loss of 3kg. The abdominal pain was continuous and non-radiated to other parts of the abdomen. He also referred meteorism, dyspepsia and abdominal distension. He had been in good health before this event. Previous medical history included cholecystectomy and right inguinal herniorrhaphy.

Physical examination revealed a soft, non-tender abdomen, with a firm abdominal painful mass in the right flank and febricula (37.4°C). Laboratory examinations were initially normal, but five days later leukocytosis (14,200 leukocytes/mm3) and high C-reactive protein levels (209 mg/dl) were found.

A 5-mm thick-slice Multidetector-row abdominal CT after administration of 120 ml of intravenous contrast material (Omnipaque™ 300mg I/ml) was performed (Fig 1, 2). CT images showed a cystic lesion with thick, irregular, enhanced walls located in the right perirenal space, among the inferior vena cava, the right kidney and the duodenum. It measured approximately 6.6 cm and showed a homogeneous liquid content without air-fluid level.

After 8 days with antibiotic treatment, the cystic lesion arising from the right perirenal space was completely excised. Escherichia Coli grew on McConkey medium. Postoperative course was uneventful.

Pathological examination of the walls of the cavity revealed a well vascularised granulation tissue with an inflammatory infiltration of neutrophile polymorphonuclear leukocytes.

Previous CT scans, MRI and ultrasound examinations showed a loculated lesion with thin walls, it suggested the diagnosis of Cystic Lymphangioma (Fig 3-5).

Discussion:

Lymphangioma is a benign lesion, with more than 80% of cases diagnosed during childhood [1]. Typical locations for cystic lymphangioma include the neck (75%) and the axillary area (20%). Cystic lymphangioma rarely occurs in the retroperitoneum or in the mediastinum (5%). Cystic lymphangiomas of the neck and the axilla are more frequently found in young children (younger than two years) while those located in the retroperitoneum are
frequently found in older children or adults [2]. The aetiology of lymphangioma is poorly understood but is generally believed to be a congenital maldevelopment. Sequestrations of foetal lymphatic sacs lack proper connections with the venous system. These blind vessels slowly proliferate and dilate, producing cystic lymphangioma [3]. Other proposed aetiologic factors are bleeding or inflammation in the lymphatic vessels [4].

Lymphangiomas are classified as simple capillary, cavernous and cystic types, depending on the size of the lymphatic spaces, the nature of the lymphatic wall and its content [5]. Cystic type, as the one we present, usually appears as a cystic, multiseptated mass with thin walls and may contain sedimentation caused by debris [5]. Only the cavernous and the cystic types have been observed in the retroperitoneum [7] and the cystic type is the most frequent intra-abdominal lymphangioma.

Intra-abdominal cystic lymphangiomas are rarely infected. Few cases have been reported, and usually the route of infection was not identified [8]. Infectious complications of these lesions which have been reported have been due to gram-positive bacteria [9] (Streptococcus pneumoniae and Staphylococcus aureus), gram-negative bacteria [8-10] (Escherichia coli, Campilobacter jejuni and Salmonella Enteritidis), and Mycobacterium tuberculosis [9].

Abdominal lymphangiomas may be the site of Salmonella seeding. Some cases of macrocystic lymphatic malformation infected with Salmonella species have been reported [9]. Macrophages may exist in lymphoid infiltrates and may constitute a potential reservoir for the survival and replication of Salmonella. It has been proposed that the route of infection could be the lymphatic drainage of the gastrointestinal tract [11].

Treatment of mesenteric cystic lymphangioma is surgical, even though total excision is required; it is not always possible because of the extensive and insinuating ramifications of the lymphangiomatous malformation [12]. Its resection may sometimes require resection of adjacent bowel or other structures because it is often closely attached to the enteric vascularity [13].

In this particular case, complete surgical excision was difficult as it was surrounding the inferior vena cava. A CT scan that was performed two months after the surgery was normal.

Differential diagnosis for cystic retroperitoneal lesions in a middle age man includes first of all cystic lymphangioma, and also cystic mesothelioma, pseudomyxoma retroperitonei, urinoma and hematoma [13]. These latter (urinoma and hematoma) can be discarded because the absence of compatible clinical history. Finally, neither the image study of 1998 nor the clinical evolution were compatible with pseudomyxoma retroperitonei or mesothelioma. So, although lymphatic tissue was not found in the wall of the cyst, it could be assumed that the lesion was an infected retroperitoneal cystic lymphangioma.

**Differential Diagnosis List:** Infected retroperitoneal cystic lymphangioma

**Final Diagnosis:** Infected retroperitoneal cystic lymphangioma

**References:**


Losanoff JE, Kjossev KT (2005) Mesenteric cystic lymphangioma: unusual cause of intra-abdominal catastrophe in


Description: Cystic lesion (C) with thick, irregular, enhanced walls located in the right perirenal space, among the inferior vena cava (V), the right kidney (K) and the duodenum (D). (A) Aorta. Origin:
Description: Cystic lesion (C) with thick, irregular, enhanced walls located in the right perirenal space, among the inferior vena cava (V). (A) Aorta. Origin:
Description: Axial T2-weighted MRI shows a retroperitoneal cystic lesion (C), located in the right perirenal space. (V) Inferior vena cava, (A) Aorta. Origin:
Description: Retroperitoneal cystic lesion, located in the right perirenal space, among the inferior vena cava, the right kidney and the duodenum. Origin:
Description: US shows a hypoechoogenic cystic lesion in the right flank without ascites. Origin: