CASE REPORT

Chronic Low Back Pain due to Retroperitoneal Cystic Lymphangioma

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Abstract

Abdominal cystic lymphangioma is a rare benign neoplasm. Less than 1% of lymphangiomas is in the retroperitoneum. Lymphangioma is mostly asymptomatic. Chronic symptoms were reported in retroperitoneal type more than others. Acute symptoms due to complications like infection, cyst rupture or hemorrhage may occur. We report an 18-years-old girl with low back pain from 6 months ago with huge pelvic mass and diagnosis of retroperitoneal cystic lymphangioma.

Key words: Low Back Pain, Lymphangioma, Retroperitoneum

Introduction

ystic retroperitoneal lesions are benign. Beahrs *et al* based on etiology categorized them into developmental, traumatic neoplastic and infectious (1). Abdominal cystic lymphangioma is very rare benign neoplasm that occurs because of problem in connection between lymphatic sac and venous system in 14-20 weeks of emberional growth (2). It can have complications or unusual presentation due to location and size. We had a case of retroperitoneal cystic lymphangioma with low back pain as a symptom. In this article we discuss the case and we review other literatures.

Case Presentation

An 18-years-old woman was referred to our department with chronic low back pain from 6 months ago. There was no history of nausea, vomiting, fever or other gastrointestinal or urinary symptoms. She had no history of surgical or medical problems. She had used pain medication for four months. In physical examination, there was lower abdominal distension and fixed large mass in the right side of the abdomen from the rib cage to the pelvis. She had no shifting dullness, tenderness or organomegaly in abdominal exam. Neurologic and musculoskeletal exam were normal . Laboratory tests showed mild normocytic anemia, while tumor markers and hydatid serology were normal. Abdominal and lumbar X-rays revealed no pathology. Due to unusual presentation of the low back pain, ultrasonography and

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computed tomography (CT) were carried out. Their report showed huge multilobulated retroperitoneal mass at right side (Figure 1). Laparotomy revealed large multi cystic lesions from liver to the pelvis with purple appearance and cysts contain fluid in the retroperitoneum in front of vertebral column associated with intestinal displacement . Mesentery of small intestine had lots of same cystic masses (Figure 2), so complete resection wasn't possible. Biopsy showed the lymphocytes and dilated lymphatic channel with cyst formation, compatible with cystic lymphangioma (Figure 3a & b). In 5 years follow –up, she had no complication or limitation for her activity and she had pregnancy with normal vaginal delivery.

Discussion

95% of lymphangioma are seen in neck and axillary region. It is reported that only 5% are observed in the abdomen including mesentery, omentum, and less than 1% in retroperitoneum. Retroperitoneal cystic lymphagioma is more common in women. 90% of cases are diagnosed before the end of second decay of life, compatible with our case was 18 years old at presentation (2). The lesions can be uniocular or multiocular like our patient. Cyst contains serous or chylous fluid(3). Lymphangioma is mostly asymptomatic and may be found accidentally during evaluation of non related clinical symptoms. Mesenteric type can cause acute abdomen because of bleeding, perforation or obstruction (4). Chronic symp-



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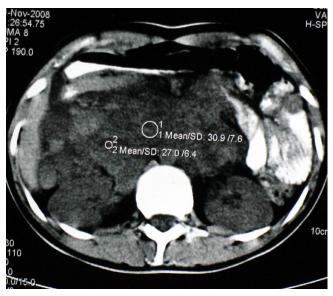


Figure 1. CT scan of 18-years-old girl with large multicystic retroperitoneal mass in front of vertebral column that caused intestinal displacement.

toms were reported in retroperitoneal type more than others. Unusual presentation may be seen. Justinger *et al* reported one case with chronic back pain like our case (5). Low back pain may occur because of giant retroperitoneal mass.

Rani *et al* reported a child with pseudoacitis because of huge mass in omentum or mesentry that has been reported as ascitis (3). Abdominal lymphangioma is divided to capillary, cavernous, and cystic type. Lymphangioma in retroperitoneum is mostly cystic. Preoperative diagnosis is possible in 25% of cases (6). Other cystic tumors of retroperitoneum from liver, kidney or pancreas may be included in differential diagnosis (7). Based on the computed tomography, we think that in our case, the mass is lymphoma or sarcoma. Ultrasound showed

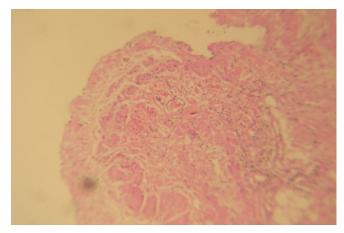


Figure 3a. Histological study shows cystic spaces covered by flat epithelium and fibrotic wall. X40 H& E stain.



Figure 2. Intraoperative picture of huge mass in right abdomen.

multiloculated cyst and CT scan reveals septated cystic mass with low enhancement. Derhy et al believed that Three-dimensional magnetic resonance lymphography is a good modality for diagnosis of retroperitoneal cystic lymphangioma. It shows that change from normal variants to lymphatic aneurysmal dilatation (8). Surgical resection is the choice of treatment in retroperitoneal lymphangioma that is possible in 55% of patients (9). Large and localized lesion can be resected completely but multiple cysts are rarely resectable and usually associate with high mortality, so organ-preserving excision of mass is needed (2, 4). Yagihashi et al and Kasza J et al reported complelete resection of unilocular retroperitoneal cyctic lymphangioma with laparoscopy (10,11) A favorite long term prognosis is expected and cure can be achieved with complete resection (12). Follow-up with ultrasound is necessary in incomplete resection to evaluate recurrence. Local injections of fibrin glue or bleomycin, laser photocoagulation or sclerotherapy with OK432 are reported as treatment modalities in retroperitoneal

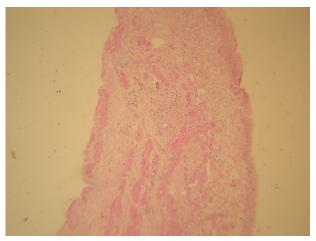


Figure 3b. Histological study shows cystic spaces covered by flat epithelium and fibrotic wall. X10 H& E stain.

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lymphangioma. Hachisuga et al reported prenatal treatment with intrauterine injection of OK432 (2).

In our case, complete resection was impossible because of extensive cyst in the retroperituneum and base of mesentery; 5 years follow- up with abdominal ultrasound did not show complications or increase in size of the mass in our patient.

Conclusion

Cystic lymphangioma is a rare lesion in adults. It may present with atypical low back pain, therefore, in some cases with atypical low back pain, other imaging modalities like ultrasonography can be helpful and lead to correct diagnosis.

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