Retroperitoneal Cystic Lymphangioma in an Aged Man: Report of a Case and Review of the Literature*

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_ Abstract —

Retroperitoneal cystic lymphangioma is a rare benign tumor which had previously been difficult to diagnose preoperatively by conventional radiographic technique(1). Recent reports describe the computed tomographic and ultrasonographic findings in cystic lymphangioma(1,2). We report a case of retroperitoneal cystic lymphangioma, accurately diagnosed by lymphography, with computed tomographic findings and percutaneous drainage findings. To the best of our knowledge, this is the most aged case of retroperitoneal cystic lymphangioma of preoperatively diagnosed.

Index Words: Retroperitoneum, lymphangioma 87.3142 Retroperitoneum, CT 87.1211 Lymphangiogram 9.125

CASE REPORT

A 65-year-old man repected dull left flank pain for 8 dyas. On physical examination, a firm tender mass was palpable on left upper lateral abdomen with mild left CVA tenderness. Computed tomography(CT) showed a huge, low-density(22.5 Hounsfield unit), cystic mass at posteromedial area to the left kidney(Fig-1), but no secondary change such as hydronephrosis is noted in the surrounding organs revealing this tumor is slowgrowing and soft consistency.

A CT guided aspiration and drainage on thid hospital day showed straw color fluid 1,000cc with protein 3.3 mg/cc and was sterile. From fifth hospital day, a milky whitish colored fluid was

drained through the previously placed drainage catheter 500-600cc daily, and showed lipid 686 mg/cc.

On the 17th hospital day, lower extremity lymphogram and lymphographic CT were done and showed a large multiseptated spaces containing droplets of iodised oil contrast medium communicating with para-aortic lymphatics at left paralumbar retroperitoneal space(Fig-2 a,b).

On the 19th hospital day, a childhead sized mass of spongy-like consistency was excised.

On pathologic report, a well demarcated tumor(weighing 72.5gm) of multiple thin-walled small cystic spaces showed microscopically dilated and degenerated lymphatic vessels with focally aggregated lymphocytes(Fig-3). The final diagnosis was cystic lymphangioma, involving

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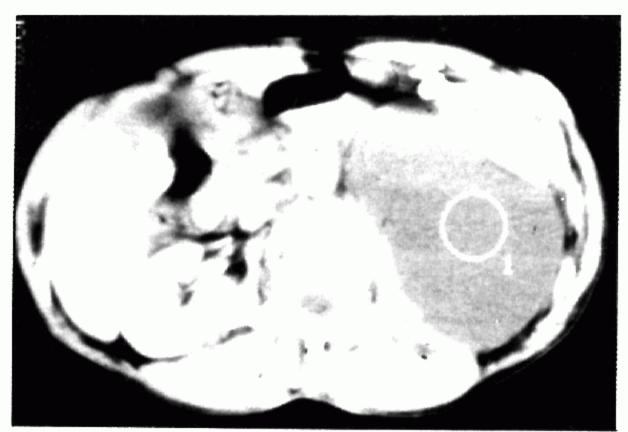


Fig. 1. Computed tomogram shows a large cystic mass in the left retroperitoneal space with 22.5 Hounsfield unit. The left kidney is markedly displaced anterolaterally, without evidence of hydronephrosis.

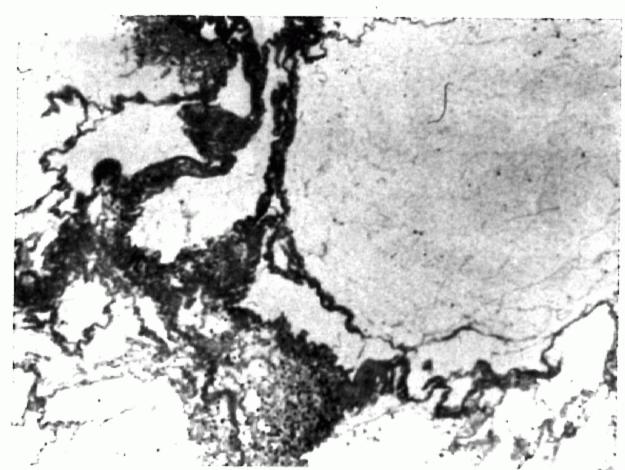


Fig. 3. Microscopic finding shows a cystic lymphangioma of dilated and degenerated lymphatic vessels and focally aggregated lymphocytes (H & E stain, x 100).

retroperitoneum.

DISCUSSION

Lymphangiomas are benign tumors of multiple dilated lymphatics. They may be classified histologically as being simple, cavernous, or cystic(3). Cystic lymphangiomas may be unilocular or multilocular and may contain serous or chylous fluid. In this case, one chamber had transudate and the other had chylous fluid as percutaneous drainage showed.

The etiology are developmental abnormality(harmatoma), trauma(lymphatic effusion and



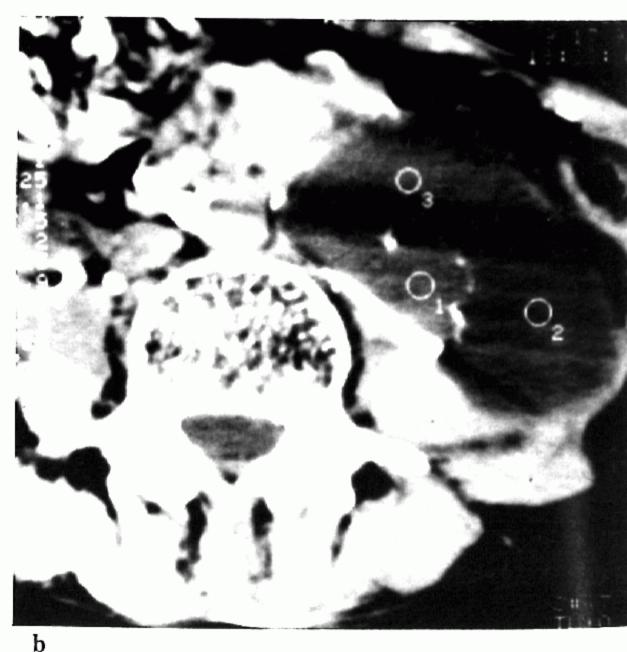


Fig. 2. Lymphogram(a) and lymphographic CT(b) show a large, multiseptated tumor mass in the left retroperitoneal space containing a few iodized of droplets of contrast media.

encapsulation) or other acquired lymphatic obstruction, but the former is the most common (1,4).

The most common site is the neck. Extremities, abdomen involving mesentery, liver or spleen, retroperitoneum and thorax were also reported (2,5-8).

Cystic lymphangiomas involving retroperitoneum most commonly present early in life as a large abdominal mass. About 50% of these lesions are present at birth and up to 90% are evident by 2 years of age(2,3,5,6). This case is very unusual in presenting symptoms later in life with a fairly benign course.

Cystic lymphangiomas seldom case acute clinical symptoms, but these may manifest themselves when the retroperitoneal cystic lymphangiomas are large enough to cause pressure on adjacent structures such as the bowel, ureters or when they are complicated by infection, hemorrhage or rupture (3,6).

On CT or ultrasonography, it is usual to find cystic areas of low attenuation value(lower than -15 HU) with multiple septa and possible bemorrhagic(1-8). But as our case, no low attenuation value cystic lesion can be found. A magnetic resonance imaging finding was also reported recently(9). Lymphogram can demonstrage a communication of the cystic mass with lymphatics as this case, but not always(3). Percutaneous thin needle aspiration was also used as a diagnostic procedure(1). As this case, the aspiration site must be carefully decised. And besides multiple punctures of different chambers are needed because the contents are variable to the punctured chamber.

As a conclusion, a cystic mass on CT with multiseptation can be a cystic lymphangioma

even though in a later in life without fat attenuation value, and lymphogram can demonstrate the communication between the mass and lymphatics and/or droplets of iodised oil contrast medium in the cystic mass.

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〈국문 요약〉

고령자에서의 후복막 낭종성임파종

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후복막강의 낭종성임파관종은 매우 희귀한 질환이며 대부분이 2세이하에서 발견된다. 저자들은 65세의 남자에서 임파조영술 및 전산화단충촬영으로 절단된 좌후복막강의 낭종성임파관종을 수술로서 확인하여 문헌고찰과 함께 보고한다. CT에서 좌후복막에 낭종성 종양을 보였으나 임파종에서의 독특한 지방조직의 Hounsfield Unit(HU)만큼 낮지는 않았고 임파계와의 관련유무를 확인할 수 없었으며, 임파조영술과 그후에 실시한 CT에서 임파관 및 임파절과의 관련성이 뚜렷하였고 임파조영제가 낭종으로 들어가 있음을 확인할 수 있었다. 따라서 고령자에서도 낭종성임파관종이 있으므로 후복막강의 낭종성 종양의 감별진단에 고려하여야 할 것이며 필요할 경우 임파관조영술을 실시해야할 것으로 사료된다.