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A Case of Retroperitoneal Lipoleiomyoma

We report a case of lipoleiomyoma which arose in retroperitoneum and presented with progressively distended abdomen in a 56-yr-old woman. The tumor was well encapsulated and consisted of two components, benign smooth muscle cells and mature adipose tissue without any atypia. It is likely to be mistaken as extrarenal angiomyolipoma, well-differentiated liposarcoma and leiomyoma with fatty change. We review the histologic characteristics of previously reported myolipoma and describe essential points of differential diagnosis.

Key Words: *Leiomyoma; Angiomyolipoma; Liposarcoma; Peritoneum*

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INTRODUCTION

Lipoleiomyoma is a benign soft tissue tumor, which was first described in 1991 (1). Since then only a few cases have been reported (2, 3). Lipoleiomyoma usually occurs within the abdominal cavity and retroperitoneum, although it may also be found in the subcutis and muscular fascia. Six cases of lipoleiomyoma have been described in the retroperitoneum (1-3), and to our knowledge, the case reported in the present study is the first in Korea. Here we report a case of retroperitoneal lipoleiomyoma and describe the histologic characteristics, and the differences of lipoleiomyoma from other tumors.

CASE REPORT

A 56-yr-old female was admitted to our hospital with progressive distension of abdomen. She had been in good health until 2 months earlier, when she noticed her increasing abdominal girth. There was no sign of tuberous sclerosis. Physical examination showed abdominal distension with palpable large mass in the left lower quadrant and mid-portion. Abdominal sonogram showed a huge, solid, heterogeneous echoic mass with multiple high echoic foci of variable size, occupying the most of abdominal and pelvic cavity. The echocharacter of the mass was consistent with lipomatous lesion. Computed tomography (CT) of abdomen and pelvis revealed a 23×21×12 cm retroperitoneal mass, extending anteriorly to the abdominal wall of left upper quadrant and inferiorly into the pelvis. The tumor displaced the small intestine to the right, the left kidney to the anterior and the uterus to the upper portion. The mass was well defined from surrounding structures and showed heterogeneous attenuation (Fig. 1). Other organs including liver, gallbladder,

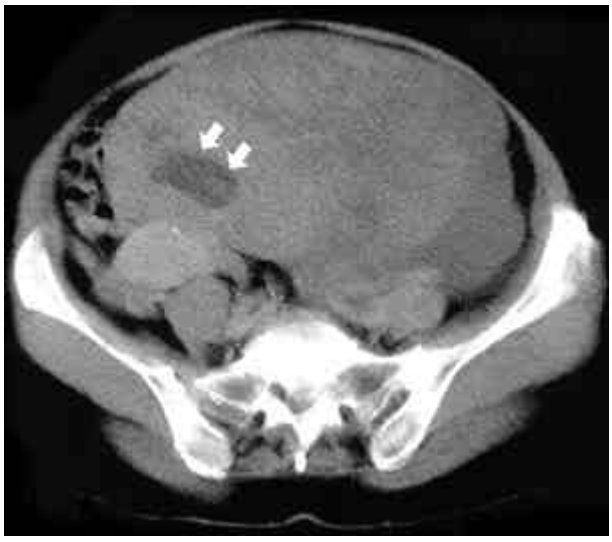

Fig. 1. Contrast-enhanced CT at level of mid-pelvis shows a huge mass with inhomogeneous attenuation. The margin is sharp. Some portions reveal low density areas (arrows), accounting for having more fat component.

Fig. 2. Cut surface of tumor is white to bright yellow with nodular whirling appearance.

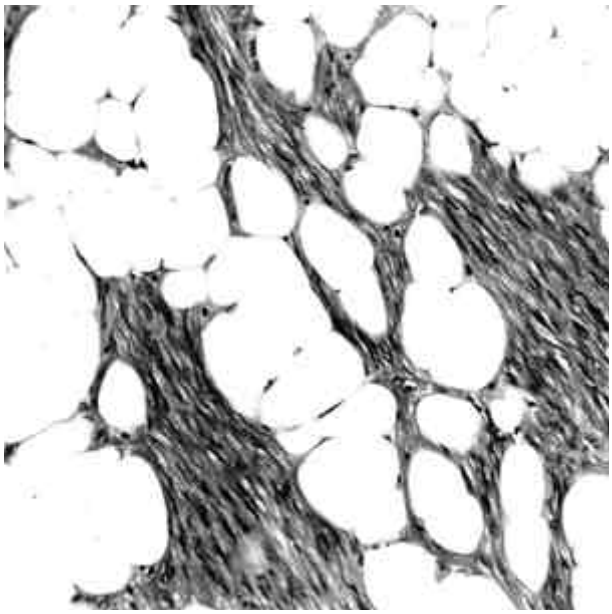

Fig. 3. The tumor is mostly composed of two components, smooth muscle cells and mature adipose cells without atypism (H&E, ×100).

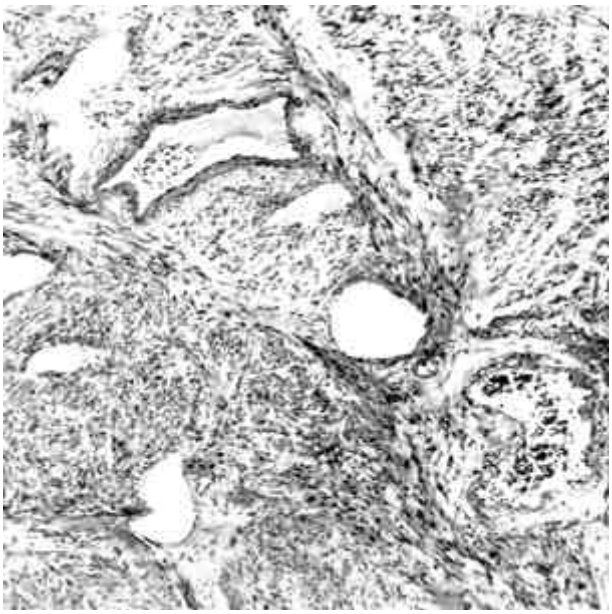

Fig. 4. Some areas show dilated and thin-walled blood vessels (H&E, ×100).

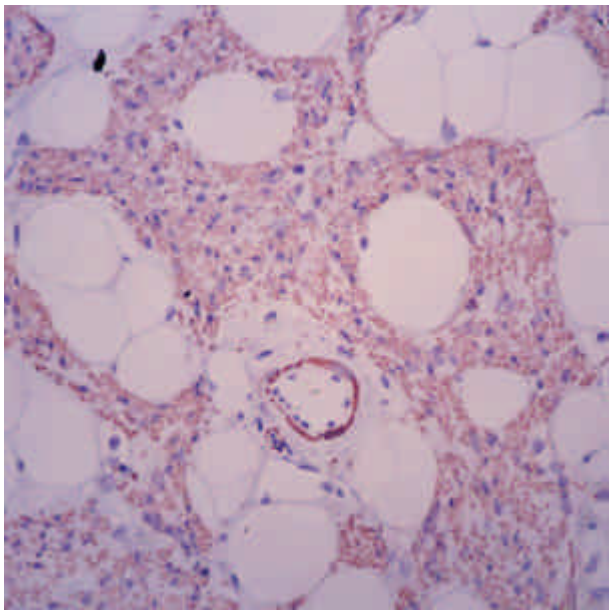

Fig. 5. Immunohistochemical staining for smooth muscle actin shows diffuse positive reaction (×200).

pancreas and spleen were unremarkable. A radiologic diagnosis of retroperitoneal liposarcoma was made. At laparotomy, a huge mass was found and resected. The tumor showed focal adhesion to the uterus and urinary bladder, but there was no infiltration to these organs. A total abdominal hysterectomy, and bilateral salpingo-oophorectomy were performed. The kidneys and liver were free of disease. The tumor excised during laparotomy measured 27×23×13 cm. The mass was well-encapsulated and lobated with rubbery to hard consistency. On sectioning, the tumor was white to bright yellow in color and lobated with nodular whirling architecture (Fig. 2). The uterus measuring 9×4.5×3 cm and bilateral adnexae were grossly unremarkable. Histologically, the tumor consisted of hypocellular spindle cells with scattered islands of fat cells (Fig. 3). The ratio of spindle cells

to fat tissue was variable depending on sampled areas, but it was approximately four to one. Dilated and thin-walled blood vessels were frequently seen (Fig. 4). Diffuse positive reaction of immunohistochemical staining for smooth muscle actin (Dako) verified that spindle cells were smooth muscle cells (Fig. 5). The spindle cells did not react with HMB-45 (Dako) and S-100 protein (Dako). The smooth muscle bundles were disposed in short interweaving fascicles and some areas showed sclerotic hyaline change. The adipose component was entirely mature without any lipoblasts. Bizarre pleomorphic cells, mitotic figures or necrosis were not present.

DISCUSSION

Myolipoma of soft tissue was first described in 1991 by Meis and Enzinger (1), of which three were retroperitoneal. These tumors show characteristic histologic finding, being composed of benign smooth muscle and mature adipose tissue. In the uterus, similar tumors are known as lipoleiomyomas, the term which some use when referring to the myolipoma of soft tissue (4). In the retroperitoneum, myolipomas could be frequently mistaken for liposarcoma, especially in radiologic diagnosis because the overwhelming majority of large retroperitoneal mass containing fat is liposarcoma (2). However, myolipomas have no lipoblasts or atypical cells and may be at least partially encapsulated, which distinguish them from liposarcoma (5, 6). Besides angiomyolipoma, spindle cell lipoma, leiomyoma with fatty degeneration, and leiomyosarcoma must be differentiated from myolipoma (1). For angiomyolipoma, it differs from myolipoma with its regular occurrence of thick, medium-sized arteries with narrow lumina and epithelioid smooth muscle cells with occasional cellular atypia, its positive reaction to HMB-45 and its frequent association with tuberous sclerosis (7-9). Spindle cell lipoma is extremely rare in retroperitoneum, and the spindle cells do not have smooth muscle differentiation. We could also differentiate lipoleiomyoma from leiomyoma with fatty degeneration. In lipoleiomyoma, adipose tissue is evenly distributed throughout the lesion, suggesting that fat is an integral part of the lesion. Furthermore, lipomatous degeneration rarely occurs in smooth muscle tumors of soft tissue. In addition, lipoleiomyoma can be distinguished from leiomyosarcoma by its bland nuclei and paucity of mitoses in the smooth muscle component.

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