# A rare case of female pelvic mass: angioleiomyoma of the broad ligament

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**Summary**

Angioleiomyoma is a benign mesenchymal neoplasm composed of smooth muscle cells and thick-walled vessels. It is usually found in the skin of the lower extremities. Angioleiomyoma is a very rare tumor among the ever-expanding repertoire of growth variants described in benign uterine leiomyoma. More rare is a solitary tumor of the broad ligament. Thus angioleiomyoma of the broad ligament is an extremely rare benign tumor of the female pelvis. In this report a 52-year-old woman with a one-year history of abdominal pain was admitted to our hospital. Gynaecological and ultrasonography exams showed a large mass with increased vascularization in the right adnexal region. The patient underwent total hysterectomy and bilateral salpingo-oophorectomy. The site of the benign mass was the left broad ligament of the uterus. On pathologic examination of the specimen, the tumor was diagnosed as angioleiomyoma. We present a case of angioleiomyoma of the broad ligament because of its extreme rarity and the large size of the tumor.

*Key words:* Female tumors; Angioleiomyoma; Broad ligament.

## Case Report

A 52-year-old menopausal woman was admitted to our department because of abdominal pain of one year's duration. The pain had a dull character and was aggravated by walking. Her body mass was stable but there were alterations of the bladder with frequency and urinary retention, constipation and abdominal distension.

On pathological anamnesis no other systemic disease was found and no surgical history was noted. Gynecological anamnesis showed gravida 2, para 2, no artificial abortion, menarche at age 12 and menopause at age 50. There was no atypical uterovaginal prolapse.

Abdominal palpation was normal but the gynecological exam revealed an uterus increased in volume, not well definable, and moved toward the right because of the presence of a fist-sized mass arising from the left side of the uterus. The mass was slightly mobile, firm and tender with an irregular outline that occupied the Douglas pouch. There was no pain on touch, the left ovary was not definable, the right ovary was enlarged in volume and touching was accompanied by pain.

Laboratory findings were all within normal range, including tumoral markers CEA, CA-125, α-FP, and β-hCG.

Ultrasonography examination performed with a pelvic and transvaginal probe confirmed the clinical evaluation and revealed a uterus measuring 170 x 71.7 x 87.6 mm displaced to the right side due to the presence of a hypoecogenic mass. Morphology showed a flattened oblong mass with a maximum longitudinal diameter of 135.5 mm detected in the left pelvis and posterior to the Douglas pouch above the uterine isthmus backward to the left.

Color Doppler scanning of the mass emphasized a color flow with regular distribution demarcating the limits of the tumor. Evaluation of the fluximetry showed the tumor had high velocity flow in the vessels with a poor resistance index.

At laparotomy the uterus, bilateral tubes and ovaries were normal. The left adnexal area was filled with a firm, round and lobulated mass that emerged from the left side of the isthmus with a soft consistency. The surface was brown and easily bled on touching. The uterus, left tube and ovary all seemed completely independent of the mass and the left ureter was not deviated. The mass was completely removed with a total abdominal hysterectomy and bilateral salpingo-oophorectomy. The postoperative period was uneventful. One year after surgery the patient is alive with no evidence of disease.

On gross examination, the left adnexal mass was a lobulated structure measuring 150 x 150 mm. It was separate and distinct from the normal left tube and ovary, emerging from the broad ligament. There was no gross evidence of malignancy (Figure 1).

The uterus measured 100 x 50 x 40 mm and showed some leiomyoma with a maximum diameter of 10 mm. The cervix was grossly normal as were the right tube and ovary.

On microscopic observation the histological report of the broad ligament mass revealed a smooth muscle tumor with poor mitotic activity, with many vascular channels and smooth muscle bundles (Figure 2).

Vascular channels were variable in size; the small-sized channels were positioned centrally and the larger ones were displaced peripherally. The small vessels were divided by fusate cells and sclerotic areas. In contrast the large vessels showed minor cell concentrations. The muscular walls of the small sized vessels were like arteries; the other ones with a thin wall were difficult to distinguish as veins.

There was nuclear polymorphism but mitotic activity was extremely rare. There was also necrosis and hemorrhagic areas, and the vascular and spindle cell components were immunoreactive for vimentin and smooth muscle for actin.

## Discussion

Leiomyoma of the uterus is common female pelvic tumor, whereas vascular leiomyoma or angioleiomyoma are rare and even more rare is a solitary tumor of the

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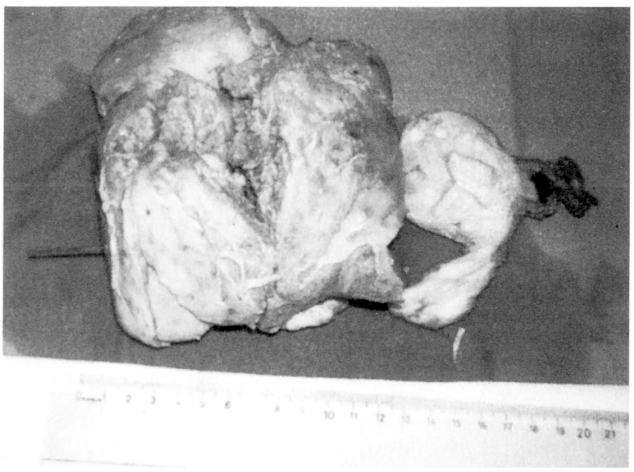

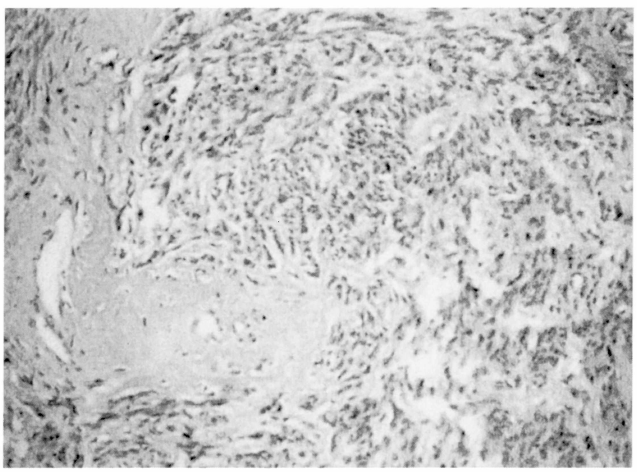

Fig. 2

Figure 1. — Gross examination of the broad ligament angioleiomyoma.
Figure 2. — Microscopic examination of the broad ligament angioleiomyoma.

broad ligament. Angioleiomyoma of the broad ligament is, therefore, an extremely rare benign tumor of the female pelvis [1].

Angioleiomyoma usually develops in the skin of the lower extremities between the fourth and sixth decades of life. It may be small and painless when located in the head and neck region [2]. However, pain is the dominant clinical feature of uterine angioleiomyoma, as in our case. The mechanism of pain is inconclusive; it may be attributable to local ischemia from vessel contraction [3].

Uterine angioleiomyoma can cause severe menorrhagia and can be difused and multiple. It has been stated that local dysregulation of the vascular structures in the uterus is responsible for abnormal bleeding. Leiomyomas contain venous plexuses, and certain growth factors have been suggested as candidates that cause abnormal bleeding [4].

On microscopic observation angioleiomyoma is composed of smooth muscle bundles which surround the vascular channels and intervene with these vessels which are closely compacted and intersecting with one another.

On the basis of histological findings the tumors have been classified into three types: capillary or solid type, cavernous type and venous type. These tumors have also been divided into two groups: a large group of extremity tumors and a smaller group of head tumors [5].

Gardner *et al.* established the definition of tumors of the broad ligament, stating that they "occur in or on the broad ligament but are completely separated from and are in no way connected with either the uterus or the ovary" [6].

According to these criteria and on the basis of pathologic findings (smooth muscle bundles surrounding the vascular channels) our case can be diagnosed as a broad ligament angioleiomyoma.

The clinical manifestations of these tumors are not specific, and include abdominal pain, palpable mass, abdominal distension, constipation, frequency and urinary retention. Spontaneous rupture of the hemoperitoneum is an emergency event [7].

The preoperative and intraoperative differential diagnosis includes any tumor that originates from or nearby the broad ligament and mimicks ovarian pathology [8]. Special stains for smooth muscle cells, such as actin, and vessel markers as CD34 and CD31 are necessary to differentiate angioleiomyoma from other neoplasms such as angiofibroma, fibroma, angiomyolipoma and angiomyofibroblastoma [7].

No cases have been diagnosed correctly before surgical intervention and only a few cases of angioleiomyoma of the uterus can be found in the literature. Hachisuga *et al.* reported 562 cases of angioleiomyoma: 500 occurrences were in the extremities, 48 in the head and 14 in the trunk; most of the tumors were less than 2 cm in diameter and none the occurrences were in the broad ligament [9].

Few cases of angioleiomyoma have been described at sites other than the extremities and head. According to Hsieh et al., the number of uterine angioleiomyomas reported was around six. Until now an occurrence on the broad ligament has not been described [10].

We have reported a rare case of angioleiomyoma of the broad ligament in a 52-year-old woman who presented with a one-year history of abdominal pain. This case is very interesting not only for the rarity of the anatomic site of occurrence, but also for the large size of the mass.

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