



Case Report

A RARE COMBINATION OF GIANT ANGIOLEIOMYOMA IN A MYOMATOUS UTERUS
AND SEROUS CYSTADENOMA OF THE LEFT OVARY

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ABSTRACT

I report a rare case of giant angioleiomyoma located in the left uterine horn of myomatous uterus and serous cystadenoma of the left ovary discovered in a 36-year-old woman. Uterine angioleiomyoma is a unique variant of leiomyoma. It usually occurs in middle-aged women and is manifested by menorrhagia, abdominal pain, and abdominal mass. Serous tumors of the ovary are the most common of epithelial tumors. Combining two benign tumors in the small pelvis is a clinical and morphological challenge. In this review, I discuss the clinical and the morphological manifestation of uterine angioleiomyoma and the accompanying diseases.

Key words:

Angioleiomyoma; Myomatous uterus;
Serous cystadenoma

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INTRODUCTION

Angioleiomyoma, otherwise known as vascular leiomyoma is a rare variant of leiomyoma originating from smooth muscle cells and containing thick-walled vessels[1]. It usually occurs in the subcutaneous tissue, most often in the lower extremities. It can rarely be located in the head and neck region the submandibular gland, and retroperitoneum. This uncommon entity has been rarely described in the female urogenital tract[2]. Angioleiomyoma is a unique lesion in the sense that there are no specific clinical and radiographic and ultrasonic features that help differentiate it from the more common leiomyoma counterpart; hence, surgical pathologists have a key role in the diagnosis, which is usually possible after a histopathologic examination[1],[2]. A full surgical incision is curative, be it as angiomyomectomy or simple hysterectomy[2].

I report a rare case of giant angioleiomyoma of the corpus uterus left horn pressed serous cystadenoma on the left ovary at a 37-year-old woman which was admitted to the hospital with severe abdominal pains, nausea, forced posture, and clinic on a sharp belly.

Case report

A 36-year-old multiparous woman visited hospital complaining of strong lower abdominal pain, nausea, and forced posture. Clinically with scars for a sharp abdomen.

Pelvic ultrasonography revealed a uterus corresponding to m.l. II, at the expense of myoma nodes. Presence of myoma node of the left uterine horn with a diameter of 24 cm. A cystic form of left ovary 4 cm in diameter with a smooth capsule. Right ovary - no pathological changes. Cavum Douglasi -no change. Paraclinical tests: leukocytes - $16.0 \times 10^9/l$; the remaining blood counts are within a normal range. Serum CA-125 in norm (26.20 U / ml).

Operational treatment was initiated at a time schedule

The laparotomy, performed under general anesthesia, confirms the uterine origin of the giant mass from the left uterine horn. The described myoma node strongly pressed the left ovary and cyst. After a complete hysterectomy and left oophorectomy, the compressed cyst captured for the left ovary is ripped. The right ovary was preserved.

Macroscopically (figure 1): A uterus with a whitish cervix and a hypertrophic endometrium. The cut surfaces of the corpus uterus fall into intramural leiomyoma nodes 1 - 3 cm in diameter (Figure 1a). Separate large tumor table with a pale greyish color and a slightly uneven smooth surface. Softly elastic on cut and with a whitish pink cut surface with areas of cystic degeneration (Figure 1b, c);no region of necrosis and hemorrhagic were found within the lesion. From the uterine fundus, the left ovary, the fallopian tube, and the ruptured ovarian cyst with brownish color are visible (Figure 1d).

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Figure a



Figure b



Figure c



Figure d

Figure 1 Macroscopic picture: a.Fundus uteri with typical leiomyomas; b.Angioleiomyoma - appearance; c. Cut surface; d.Fundus uteri, left ovary with a fallopian tube and torn serous cystadenoma.

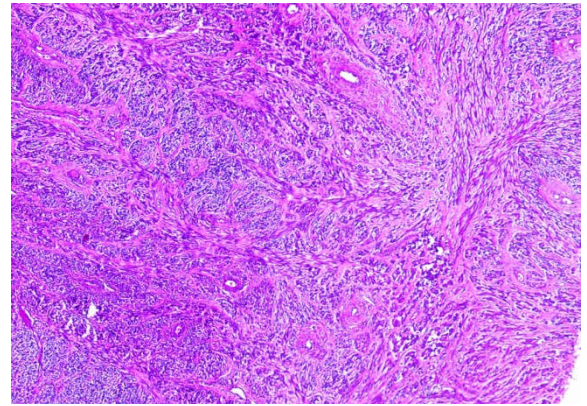


Figure a

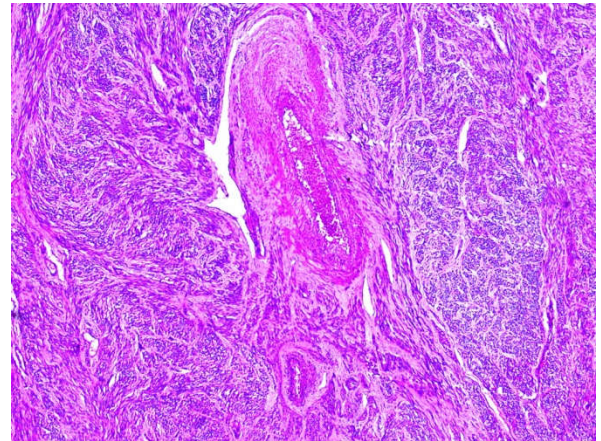


Figure b

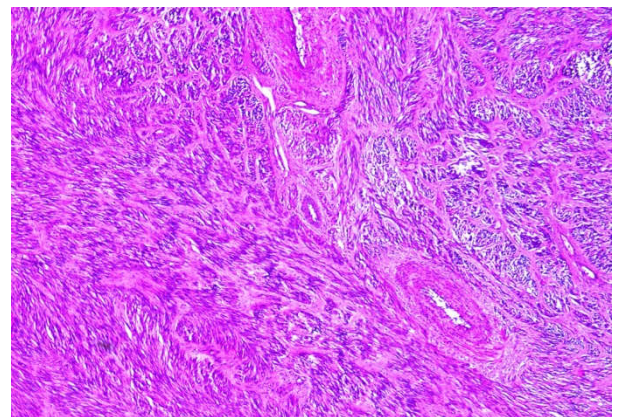


Figure c

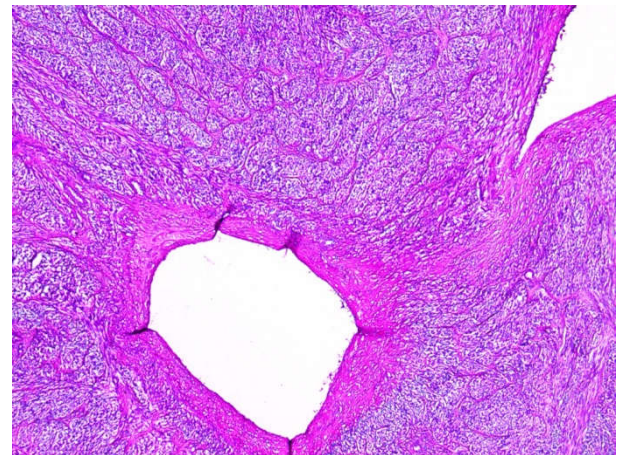


Figure d

Figure 2 Microscopic images of uterine angioleiomyoma (H&E): a,b,c.The giant nodule shows a moderately cellular spindle cell tumor composed of interlacing fascicles of spindle to plump cells with interspersed abundant thick-walled muscular vessels(enlargement $\times 50$); d.blood vessels from venous type(enlargement $\times 100$)

Histology Hematoxylin and Eosin (H&E) (figure 2): The tumor was composed of interlacing fascicles of smooth muscle cells with prolonged nuclei and hyaline fibers with interspersed abundant thick-walled blood vessels with a different caliber (Figure 2a and c) and blood vessels from venous type (Figure 2d), are missing necrotic fields and mitotic figures. Corpus uterus with hypertrophic endometrium. Myometrium with adenomyosis. Sections from the other intramural nodules showed classical histomorphology of conventional uterine leiomyomata. Fallopian tube with fibrosis and endometriosis in the wall. Left ovary with corpus albicans and corpus luteum. Follicular cysts and a section with endometriosis. Serous cystadenoma with massive hemorrhages in the wall and in a single area with stored cubic epithelium.

Immunohistochemistry (Dako, Glostrup, Denmark): the tumor positive for Desmin, SMA, Vimentin. Negative for p53, HMB45, and Ki67proliferative activity, not more than 5%. Based on the above findings and immunohistochemistry, the tumor was diagnosed as a benign angioleiomyoma.

Diagnosis: Angioleiomyoma in a myomatous uterus. Adenomyosis. Serous cystadenoma of the left ovary. Endometriosis of the left fallopian tube and left ovary.

DISCUSSION

Angioleiomyoma of the uterus is a very rare benign tumor, originating from the mesenchymal tissue and composed of smooth muscle cells and thick walled vessels[1]. Apart from the uterus, localizations are rare and in the retroperitoneum, broad ligament, and ovary[3],[4],[5]. The reported case is the first angioleiomyoma with uterine horn localization and with such a huge size. This localization with the concomitant existence of serous cystadenoma on the left caused the cystadenoma to be pressed and provoked acute abdominal symptoms -abdominal pain, nausea, forced posture, and peritoneal irritation. The clinic, echographic image, and elevated leukocytes necessitated an urgent hysterectomy. Two days after hysterectomy, leukocytes are within a normal range. Histologically, angioleiomyoma can be classified into 3 subtypes: capillary or solid, cavernous, and venous. They occur in the myometrium and usually have a size of up to 4 cm. They are usually encapsulated, multi-loculated and contain numerous vessels[6]. In the reported case we have leiomyomas in the uterine body and giant angioleiomyoma in the left uterine horn. Histologically, a number of solid and rare vessels of capillary and venous type. The preoperative diagnosis of angioleiomyoma is extremely difficult or even impossible and can be diagnosed after surgical procedure and histological assessment by involving immunohistochemical staining. In the case reported, is established before surgery: a myomatous uterus, a large nodule in the left uterine horn and a cyst beside it. Leading was the clinic of a sharp belly. Macroscopically: the tumor was more like a sarcoma with its characteristic appearance, but there were no bleeding or necrosis.

A series of slices through 3 microns was performed to establish multiple vessels, collagen fibers and cells without atypia. Negative HMB45, p53, and low Ki67 reject the possibility of leiomyosarcoma and Perivascular epithelioid cell tumor. The case is puzzled by the combination of two benign tumors in a small pelvis, as the solid tumor presses the cystic firmly and provokes a sharp abdomen. From the case presented and from other reports[1],[2],[6], it is established that angioleiomyoma has no particular imaging findings to differentiate it from other smooth muscle neoplasm. Uterine angioleiomyoma has a preoperative differential diagnosis, which is difficult from other tumors. Therefore, it is important for the clinicians and pathologists to recognize this rare benign tumor and differentiate it from similar tumors, including malignant.

CONCLUSION

The uterine angioleiomyoma is a rare benign variant of uterine leiomyoma composed of smooth muscle cells and thickened vessels. Preoperative diagnosis is very difficult and a definite diagnosis can be made only after histopathologic examination. Depending on its location and combination with other processes it can provoke various symptoms. The complete surgical excision such as angiomyomectomy or simple hysterectomy is an effective treatment.

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