Multiple uterine angioleiomyomas mimicking an ovarian neoplasm: a case report

S. Hakverdi[1], *M.D.*; K. Dolapçioğlu[2], *M.D.*; A. Güngören[2], *M.D.*; M. Yaldiz[1], *M.D.*; A.U. Hakverdi[2], *M.D.*

[1]Department of Pathology, [2]Gynecology and Obstetric Surgery, Faculty of Medicine, Mustafa Kemal University Antakya (Turkey)

Summary

Angioleiomyoma is a rare benign neoplasm that originates from smooth muscle cells and contains thick-walled blood vessels. A 69-year-old woman had been complaining of progressive abdominal distention for the last five years. At the exploration, a tumoral mass that could not be differentiated as to whether it had its origin from the uterus or ovary was specified. The patient underwent total hysterectomy and bilateral salpingo-oophorectomy (TAH+BSO). On pathologic examination of the specimen, the tumor was diagnosed as an angioleiomyoma. Here, we present a case of giant-multiple uterine angioleiomyomas mimicking an ovarian neoplasm.

Key words: Angioleiomyoma; Uterus.

Introduction

Angioleiomyoma is a benign mesenchymal tumor composed of smooth muscle cells and thick-walled vessels. It is usually found in the skin of the lower extremities. There have only been a few cases of uterine angioleiomyoma reported in the available English literature. We present a case of giant-multiple uterine angioleiomyomas causing progressive abdominal distention.

Case Report

A 69-year-old patient had been complaining of progressive abdominal distention for the last five years. She had hypertension and was using antihypertension drugs. At her first physical examination there was an immobile left deviated growth, a huge mass 2-3 cm above the umblicus which could not be clearly differentiated as to whether it originated from the uterus. Laboratory findings were within normal limits, including tumor markers CEA and CA-125. At abdominal magnetic resonance imaging (MRI) examination, the mass was nearly 19 x 16 x 15 cm in size and had a complex structure and thick walls. The multicystic tumoral mass originated from the left ovary. At the middle of the mass, the uterus was found to be 16 x 11 x 7 cm in size containing multiple myomas. There was no free fluid or LAP in the abdominal cavity and Dougla's space. Breast and gastrointestinal system examinations were normal. According to these results, it was decided to perform laparatomy for the uterine myomas and ovarian tumor. At exploration, the size of the uterus was approximately 16 weeks' gestation with a myomatosis structure. There was an approximately 20 x 16 cm in size tumoral mass that could not be differentiated as to whether it had its origin from the uterus or ovary because of tight adherence to each other, and the left ovary could not be seen. It was decided to carry out TAH + BSO. At the operation after dissection of the bladder from the uterus, a 5-6 cm cervical myoma was found originating from the right anterior wall of the cervix. Surgery was completed without complications. The patient was discharged three days later.

At gross examination, the hysterectomy specimen weighed 4500 g and was lobulated. Both the intramural and subserosal tumors measured 19 x 13.5 cm, and an additional tumor next to the ovary was 7 x 4 cm in size. The cut surface was whitish-yellow in color with punctuated hemorrhage (Figure 1).

Microscopic examination showed that all the masses in the myometrium and next to the ovarian masses were composed of interlacing smooth muscle bundles, without significant pleomorphisim, intermingled with abundant thick-walled vessels (Figure 2). Small areas of myxoid degeneration were also found in many sections. There were no mitotic figures or necrosis. Spindle cell components were immunoreactive for vimentin and smooth muscle actin and the vascular component was immunoreactive for CD34 (Figure 3). The final histopathologic diagnosis was angioleiomyoma.

Discussion

Angioleiomyoma is a rare benign soft tissue tumor that originates from smooth muscle cells and contains numerous thick-walled blood vessels [1,2]. It usually occurs in the subcutis and prefers the extremities, particulary the lower leg [1]. It can also be located in the head and neck regions [3]. Few cases of angioleiomyoma have been described at sites other than the extremities and head. The female genital tract is rarely involved. There were only 11 cases of uterine angioleiomyoma reported in the available English literature from 1966 to 2007 [4-7]. We found one previous case report of multiple huge angioleiomyomas of the uterus causing severe menorrhagia [4]. Our case is the second multiple huge angioleiomyomas of the uterus mimicking an ovarian neoplasm.

There are several uncommon specific types of leiomyomas. However, the karyotype evaluation did not fit the classical subgroups of an ordinary uterine leiomyoma [8].

Angioleiomyoma develops later in life, usually between the fourth and sixth decades [1]. It may be small and painless when it is located in the head and neck

Revised manuscript accepted for publication August 18, 2008

Fig. 1

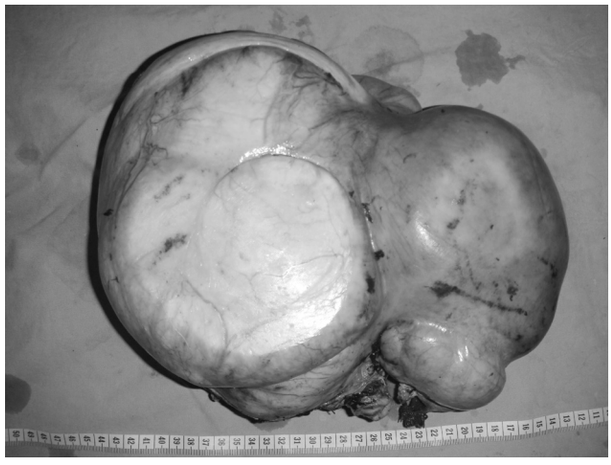

Fig. 2

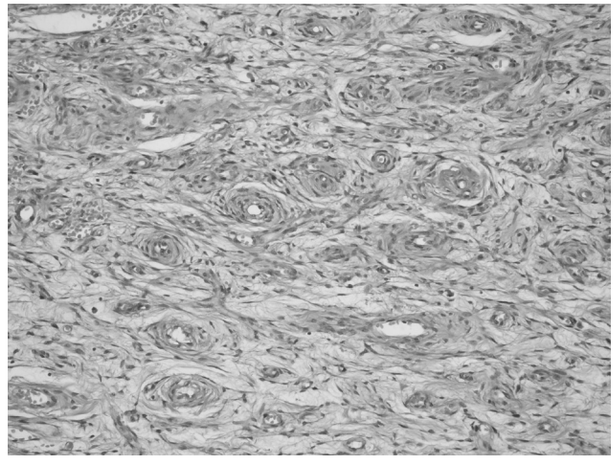

Fig. 3

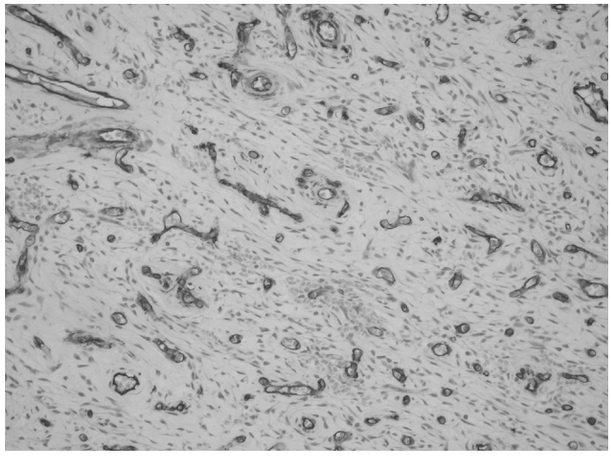

Figure 1. — Gross appearance of the uterus containing the neoplasm.

Figure 2. — Microscopic examination reveals numerous thick-walled vessels interspersed between smooth muscle cells (original magnification, hematoxylin x 200).

Figure 3. — Photomicrograph examples of CD34 staining by immunohistochemistry in the thick-walled vessels (original magnification, CD 34 x 200).

region [3]. However, pain is the dominant clinical feature of uterine angioleiomyoma causing intraabdominal bleeding by rupture [5]. It can cause severe menorrhagia and can be huge and multiple [4]. In our case, there was a palpable mass and abdominal pain.

Grossly, the tumors are circumscribed, gray-white nodules. Sometimes, the tumoral mass can have dilated vessels that can be mistaken for a multiloculated and multiseptated ovarian neoplasm as in our case.

Microscopically, the well demarcated nodule was composed of whorled, fusiform smooth muscle cells with thick-walled vessels. Areas of myxoid changes, hyalinization, calcification, and fat could be seen. There were no mitotic figures or necrosis. Angioleiomyomas are classified into three histologic types: capillary or solid, cavernous, and venous [4]. Classification is based on the variable relationship between smooth muscles and vascular cavities of different shapes.

Clinical diagnosis may be difficult, especially when degenerative changes occur. When there is heavy bleeding, sometimes it can be mistaken for ectopic pregnancy or a malignant gynecological tumor [5].

It can be difficult to distinguish angioleiomyomas from hemangiomas or an arteriovenous malformation if the vascular component predominates. Angioleiomyomas are well circumscribed neoplasms that contain at least foci of typical spindled smooth muscle cells [2]. Hemangiomas of the uterus are rare localizations for this type tumor, and are usually cavernous type. Hemangioma and arteriovenous malformations tend to be poorly defined grossly and microscopically [9]. Special stain for smooth muscle cells, such as actin, and vessel markers as CD 34 and CD 31 are necessary to differentiate angioleiomyoma from other neoplasms such as fibroma, angiofibroma, or angiomyofibroblastoma.

The main treatment for angioleiomyoma is complete excision. Either angiomyomectomy or simple hysterectomy for women who do not wish to have more children has proved to be an effective treatment. Of the reported 11 cases and our one case with uterine angioleiomyoma, hysterectomy was performed in 11 and angiomyomectomy was performed in one patient.

In conclusion, uterine angioleiomyoma should be taken into account in the differential diagnosis of a multilobule mass located in the pelvis, and as in our case, it may mimic an ovarian neoplasm. Both angiomyomectomy and simple hysterectomy have proven to be effective treatments in these cases.

References

[1] Weiss S.W., Goldblum J.R.: "Benign tumors of smooth muscle". In: Enzinger and Weiss's Soft Tissue Tumors. 4th edition, St. Louis, Mosby, 2002, 699.

[2] Zaloudek C., Hendrickson M.R.: "Mesenchymal tumors of the uterus". In: Kurman R. (ed.), Blaustein's Pathology of the Female Genital Tract. 5th edition, New York, Springer-Verlag, 2002, 574.

[3] Wang C.P., Chang L.Y., Sheen T.S.: "Vascular leiomyoma of the head and neck". *Laryngoscope,* 2003, *114*, 661.

[4] Hesieh C.H., Lui C.C., Huang S.C., Ou Y.C. *et al*.: "Multiple uterine angioleiomyomas in a woman presenting with severe menorrhagia". *Gynecol. Oncol.,* 2003, *90,* 248.

[5] Culhacı N., Ozkara E., Yüksel H., Ozsunar Y., Unal E.: "Spontaneously ruptured uterine angioleiomyoma". *Pathol. Oncol. Res.,* 2006, *12,* 50.

[6] Cobellis L., Pecori E., Rigatti F., Scaffa C., Rotondi M., Messalli E.M.: "A rare case of famela pelvic mass: angioleiomyoma of the brad ligament". *Eur. J. Gynaecol. Oncol.,* 2007, *28*, 418.

[7] McCluggage W.G., Boyde A.: "Uterine angioleiomyomas: "a report of 3 cases of a distinctive benign leiomyoma variant". *Int. J. Surg. Pathol*., 2007, *15*, 262.

[8] Mastilovic K., Rajovic J., Zigic D., Stojiljkovic B.: "Hemangioma of the uterus". *Arch. Oncol.,* 2005, *13,* 148.

[9] Hennig Y., Caselitz J., Stern C., Bartnitzke S., Bullerdiek J.: "Karyotype evaluation in a case of uterine angioleiomyoma". *Cancer Genet. Cytogenet.,* 1998, *108,* 79.

Address reprint requests to:
S. HAKVERDI, M.D.
Bacelievler Sitesi Kardelen sok no. 3
31030 Antakya/Hatay (Turkey)
e-mail: hakverdisibel@yahoo.com