Case Report

Symptomatic Primary Ovarian Leiomyoma in a Postmenopausal Woman: A Rare Entity

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Abstract

Leiomyomas are benign mesenchymal neoplasms mostly seen in the uterus and are one of the most common pelvic masses seen in women, but primary ovarian leiomyomas are rare among all the benign ovarian tumors, which account only for 0.5%–1%. The definitive diagnosis of such lesions is difficult prior to surgical excision, as there are no pathognomonic symptoms or characteristic imaging findings. Here, we report a case of primary ovarian leiomyoma with brief review of literature, highlighting the differential diagnosis of ovarian spindle cell lesions. The correct diagnosis of an ovarian leiomyoma requires identification of the nature of tumor as smooth muscle. An immunohistochemistry marker analysis is recommended for definitive diagnosis.

Keywords: *Leiomyoma ovary, postmenopausal, spindle cell lesions*

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Introduction

Among all the benign primary ovarian smooth muscle tumors, ovarian leiomyoma is uncommon, accounting for 0.5%–1% of all tumors.[1] Approximately seventy cases have been reported in the literature. Most of these tumors are unilateral, are small in size, are usually <3 cm in size, and generally occur in premenopausal women, and postmenopausal cases account for approximately 16%.[2] Patients are usually asymptomatic, and the tumor is most commonly diagnosed incidentally by histopathological examination of ovarian tissue after an oophorectomy for solid ovarian mass. Ovarian leiomyoma probably arises from the smooth muscle cells of the blood vessels present at hilum of the ovary, but other possible sites of origin are from smooth muscle cells in ovarian ligaments, multi-potent cells in the ovarian stromal tissue, undifferentiated germ cells and cortical smooth muscle metaplasia. Tomas *et al*. had suggested that ovarian fibroids could arise from smooth muscle metaplasia of endometriotic stroma or myofibroblast of ovarian stromal cells.[3] Ovarian leiomyomas are concomitantly seen with uterine leiomyomas (78%) that suggests stimulation by an identical hormone.[4] Because ovarian leiomyomas are rare and have gross and histological similarities with other spindle cell tumors such as thecoma/fibroma, sometimes, the diagnosis can be difficult.[5] Postmenopausal women presenting with large solid ovarian tumor raises the suspicion of malignancy.

Case Report

A 68-year-old postmenopausal woman presented with the complaint of abdominal pain for 1 month. Per abdominal examination revealed the presence of tenderness in the right iliac region. Complete blood count and laboratory parameters were normal. Her serum CA-125, CA-15-3, CA-19-9, carcinoembryonic antigen, and alpha fetoprotein values were within normal limits. Transvaginal pelvic ultrasound revealed a right adnexal mass of size 6.6 cm × 6.2 cm × 6.1 cm, with an irregular external surface that showed a heterogeneous isoechoic pattern. During surgery, inspection of the uterus and adnexa revealed a solid, firm, right-sided ovarian tumor with a nodular surface measuring 8.5 cm × 7.2 cm × 6.8 cm. The tumor was distinctly separated from the uterus and exhibited no adhesion or infiltration of the surrounding structures, and there was no accompanying uterine mass. The left Fallopian tube

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and ovary were normal on inspection and were left intact. On gross examination, we received a formalin-fixed uterus cervix with right-sided adnexa measuring 8.5 cm × 4.5 cm × 3 cm. The uterine cervix was found to be normal, and on serial sectioning, no fibroid was identified in the uterus. The right ovary was measuring 8.5 cm × 7.2 cm × 6.8 cm. The external surface was nodular and hard; cut surface was gray-white solid with whorled appearance, with peripheral compressed ovarian tissue having corpus luteum [Figure 1a and b]. The right Fallopian tube was measuring 5 cm in length, and the lumen was patent. Microscopically, cervix, endometrium, myometrium, and right Fallopian tube were unremarkable. However, the ovary showed a rim of normal ovarian stroma having corpus luteum along with a lesion component comprising of intersecting and interlacing fascicles of spindle-shaped cells arranged in a whorled pattern, with minimal pleomorphism. The spindle cells had blunt-ended nuclei and moderate amount of eosinophilic cytoplasm [Figure 1c]. There were extensive areas of edema and hyaline degeneration. Mitotic activity was sparse, 0–1/10 high-power field. Necrosis was not observed. Based on these features, the tumor was diagnosed as benign ovarian leiomyoma. Staining with Masson's trichrome showed pink-colored smooth muscle fibers separated by blue-colored fibrous tissue [Figure 1d]. On immunohistochemistry, the tumor showed intense positivity for smooth muscle actins and positivity for h-Caldesmon and was negative for calretinin and inhibin [Figure 1e and f]. Ki-67 proliferation index was low <2%. The final diagnosis of primary ovarian leiomyoma was given. Like uterine smooth muscle tumors, same criteria are applied for ovarian leiomyoma. The postoperative period was uneventful.

Discussion

Most ovarian leiomyomas described in literature are asymptomatic and are discovered either during routine physical examination, incidentally at surgery, or at autopsy.[6] In symptomatic cases, clinical presentations are variable such as abdominal pain, a palpable mass, hydronephrosis, elevated CA-125, polymyositis, hydrothorax, and ascites.[7] Uterine leiomyomas and adenomyosis have been diagnosed concomitantly with ovarian leiomyoma by some authors. Ovarian leiomyomas show hyaline degeneration; myxomatous changes have also been reported.[3] In our case, symptomatic primary ovarian leiomyoma was presented with secondary degenerative changes without uterine leiomyoma and adenomyosis in a postmenopausal woman. Primary ovarian leiomyoma is often misdiagnosed preoperatively as pedunculated uterine myoma, ovarian fibroma, or even ovarian endometrioma.[8] The present tumor showed a rim of normal compressed ovarian tissue, thereby establishing the primary nature of the lesion, the smooth muscle origin of which was confirmed on immunohistochemistry (IHC). The major differential diagnosis for ovarian leiomyoma includes

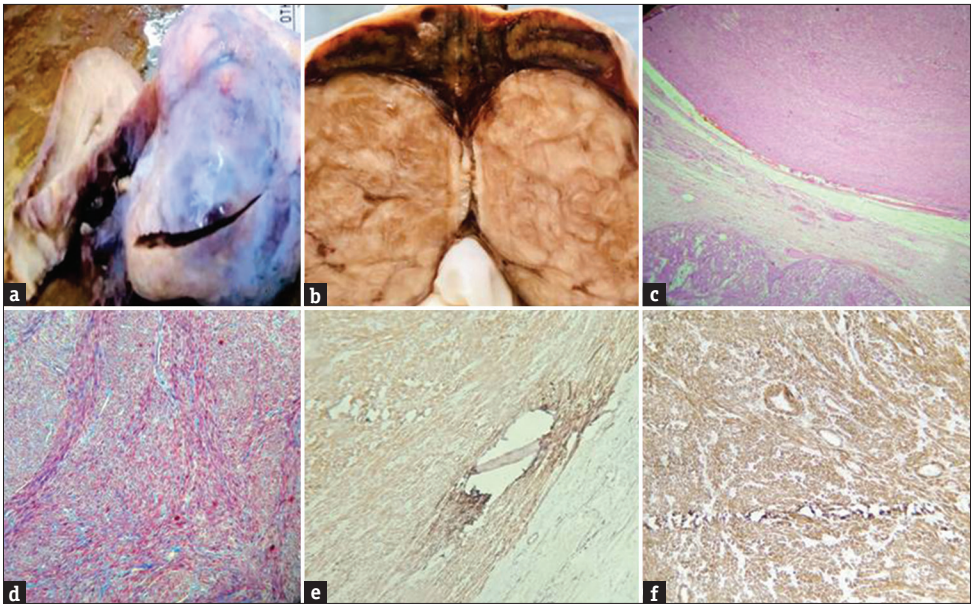

Figure 1: (a) Uterus cervix with the right adnexa showing external nodular surface of the ovarian mass. (b) Cut surface of mass show gray-white whorled appearance with peripherally compressed ovarian tissue showing corpus luteum. (c) Fascicles of smooth muscle cells and corpus luteum (H and E, ×40). (d) Pink color interlacing bundles of smooth muscle cells separated by thin, delicate blue collagen fibers (Masson's trichrome, ×100). (e) Strong cytoplasm immunoreactivity for h-Caldesmon (×40). (f) Diffuse strong cytoplasmic and membranous immunoreactivity for smooth muscle actins (×100)

leiomyosarcoma and sex cord-stromal tumors such as ovarian fibroma/thecoma, cellular fibroma/thecoma, and sclerosing stromal tumor. Although leiomyomas are easily diagnosed on H and E alone, in view of the age of the patient and location, IHC markers were applied to rule out other differentials. On IHC study, leiomyoma shows a strong positivity with α-smooth muscle actins and diffuse positivity with desmin, thus differentiating it from other spindle cell tumors of ovary. Leiomyoma also stains with Masson's trichrome and caldesmon.[9] Fibroma although show positivity for α-smooth muscle actins, but negative or focal positive for desmin, thus helping in its differentiation from ovarian fibroids. Cellular thecoma does not express smooth muscle actins and instead expresses α-inhibin and calretinin.[3] In case of huge tumors, malignancy should be ruled out despite its benign appearance on microscopy, so the ovarian leiomyomas must also be differentiated from leiomyosarcoma by using the criteria of mitotic count, cytological atypia, and tumor necrosis.[3] In our case, none of these criteria were met. For confirming mitosis, the Ki-67 proliferation index was assessed and found to be <2%. Ki-67 is an index protein that affects growth control in leiomyoma monoclonal cells and is helpful in the evaluation of characteristics of neoplastic process of uterine leiomyoma.[10] Spindle cell carcinoma and metastatic gastrointestinal stromal tumor should also be excluded in case of large tumors. Ovarian leiomyomas have a benign course and histopathology, as well as IHC confirms the diagnosis. Complete surgical resection is the preferred treatment. This is an additional case of ovarian leiomyoma in a postmenopausal woman.

Conclusion

Primary ovarian leiomyomas are rare tumors of unresolved origin and should be considered in the differential diagnosis of ovarian spindle cell tumors. Preoperative diagnosis can be difficult with solid ovarian tumors in some cases and requires additional IHC study for definitive diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent form. In the form the patient has given her consent for her clinical information & images to be reported in the journal. The patient understands that her name & initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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