Primary Ovarian Leiomyoma—A Common Tumor at an Uncommon Site

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Abstract

Leiomyoma is a benign mesenchymal tumor that commonly occurs in the uterus, ovary being a rare site. Ovarian leiomyomas constitute 0.5 to 1% of all benign tumors. They probably arise from the smooth muscle cells in the ovarian hilar blood vessels. They occur most commonly in premenopausal women. We report a case of primary ovarian leiomyoma in a 39-year-old female patient.

Keywords
- leiomyoma
- ovary
- smooth muscle tumor
- histopathology
- Masson's trichrome stain

Introduction

Leiomyoms are extremely common neoplasms, overall incidence being 77%. It most commonly occurs in women older than 50 years.[1] The most common site is the uterus. Other less common sites are cervix, uterine ligaments, and ovary. Primary ovarian leiomyoma is a very rare tumor and it accounts for 0.5 to1% of all the benign ovarian neoplasms. They probably arise from smooth muscle cells in the ovarian hilar blood vessels.[2] They occur most commonly in premenopausal women. Majority of these tumors remain asymptomatic and are detected incidentally during physical examination or surgery. Most of these tumors are unilateral. However, in the pediatric and young adult group they are more commonly bilateral. No bilateral cases have been described in patients older tha35 years in literature.[3] The most useful modalities for detecting extrauterine leiomyomas are ultrasonography (USG), computed tomography (CT), and magnetic resonance imaging (MRI).[4] Surgical excision is the treatment of choice.[5]

We report a case of primary ovarian leiomyoma in a 39-year-old woman presenting with pain in the abdomen and abdominal distension.

Case History

A 39-year-old patient came to obstetrics and gynecology department of tertiary care hospital with complaints of pain in the abdomen and abdominal distension since 2 months. Pain in the abdomen was in the form of dull aching and intermittent type in the right lower abdomen with gradual abdominal distension. She also gave a history of weight loss and irregular periods since 2 months. Per abdomen examination, an ill-defined mass was noted corresponding to 20 weeks in size, firm in consistency, and mobile. Vaginal examination revealed a mobile mass, separate from the uterus. Her routine blood tests were within normal limits. USG abdomen and pelvis showed a 12 ×12 cm, large heterogenous, hypoechoic, solid mass in the right adnexa with minimal vascularity. Uterus was displaced toward left. A small cystic component was seen along the periphery of the anterior aspect of the mass, probably ovarian tissue. Left ovary and uterus appeared to be normal. Minimal ascites was noted. A probable diagnosis of a large broad ligament fibroid and solid ovarian fibroma was made.

Laparotomy with total abdominal hysterectomy with bilateral salphingo-oophorectomy (TAH+BSO) was done. Intraoperatively, a right-sided ovarian mass was seen, 12 × 12 cm in size with bosselated surface, firm in consistency. Broad ligament was free and the mass was mobile (►**Fig. 1**).

Left ovary and bilateral tubes appeared to be normal. Free fluid was present in the peritoneal cavity. Peritoneal washing was sent to cytology which was negative for malignant cells.

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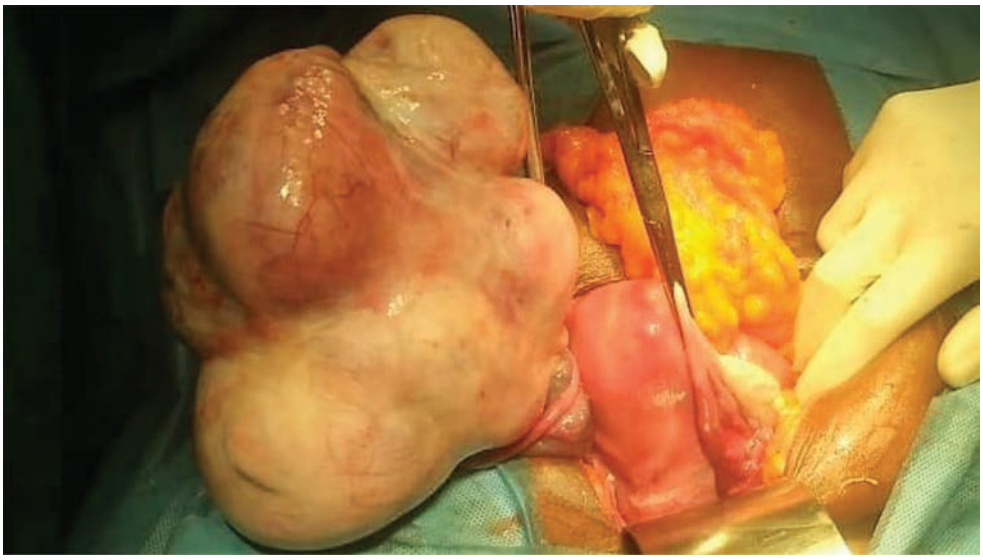

Fig. 1 Intraoperative picture showing the mass arising from right ovary. Broad ligament is free.

Grossly, we received TAH+BSO specimen. Uterine myometrium showed a small intramural fibroid measuring 0.4 × 0.4 cm. Right ovary could not be separately visualized. A well circumscribed mass was seen below the right fallopian tube measuring 17 ×12.5 cm. Outer surface appeared congested with nodular, bosselated appearance. The cut surface of the mass showed solid, yellowish to pale white, whorled appearance (►**Fig. 2**).

Further search for right ovary revealed a nodule adjoining the mass with cystic consistency, cut surface of which oozed serous fluid. Cyst wall was yellowish and smooth. Compressed ovarian tissue was noted adjacent to the cyst and the mass.

Left ovary showed corpus albicans and a corpus luteal cyst. Bilateral tubes appeared normal. Microscopically, right adnexal mass showed smooth muscle cells arranged in interlacing bundles and fascicles suggesting leiomyoma (►**Fig. 3**).

The diagnosis was substantiated by Masson's trichrome stain (►**Fig. 4**).

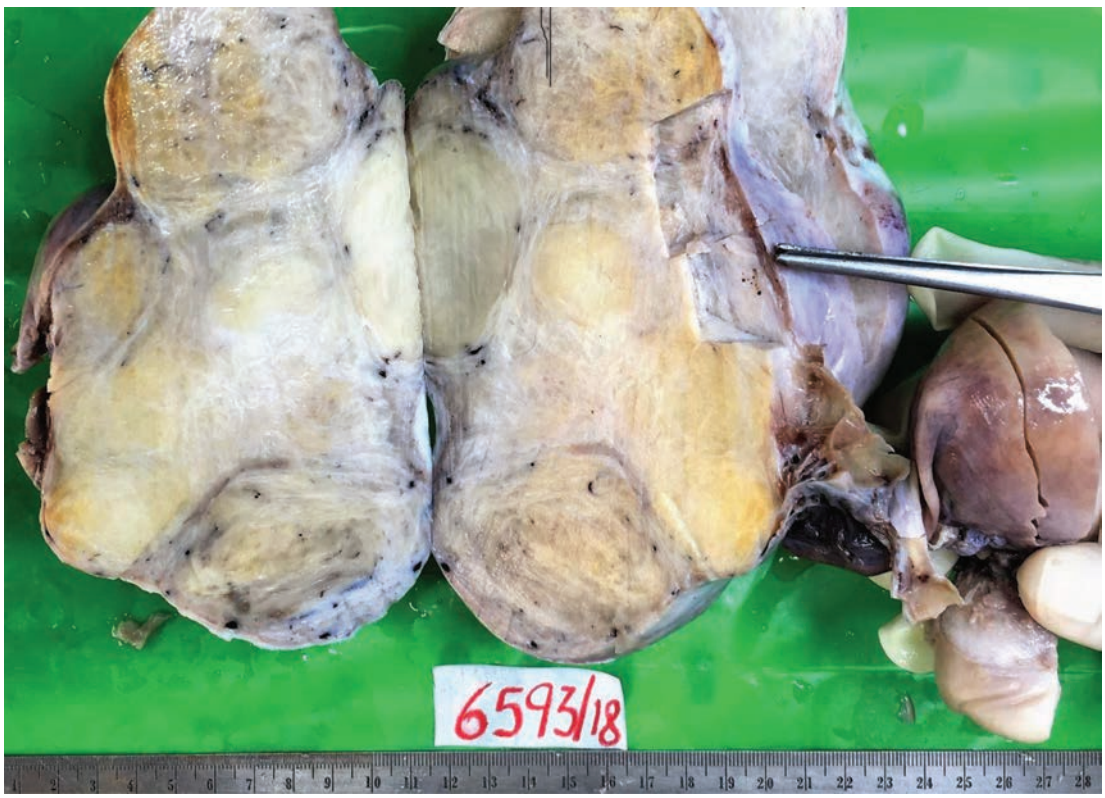

Fig. 2 Total abdominal hysterectomy specimen, cut surface of right ovarian leiomyoma with whorled appearance. Compressed ovarian tissue pointed by forceps.

Sections from the compressed ovarian tissue showed ovarian stroma and hilar vessels at the periphery of the mass (►**Figs. 5** and **6**).

Bilateral tubes were of normal morphology. Left ovary showed corpus luteal cyst. Intramural fibroid showed features of leiomyoma. Final diagnosis of right ovarian leiomyoma was made.

Discussion

Primary ovarian leiomyoma is a very rare benign smooth muscle tumor. Since its first description in 1862 by Sangalli et al, only 70 cases of this rare tumor have been reported in the literature.[3]

The histogenesis of ovarian leiomyoma remains uncertain. Several theories have been proposed regarding its potential origin. Few studies state that they probably originate from smooth muscle cells in ovarian hilar blood vessel, cells in the ovarian ligament, smooth muscle cells or multipotential cells in the ovarian stroma, undifferentiated germ cells or cortical smooth muscle metaplasia of endometriotic stroma, smooth muscle present in mature cystic teratomas, and smooth muscle in the wall of mucinous cystic tumor. They often coexist with uterine leiomyoma suggesting a common etiology or identical hormonal stimulation.[1]

Primary ovarian leiomyomas most commonly occur in premenopausal women; however, it has a wide age distribution of 20–65 years.[3] Most of the times, patients are mostly asymptomatic. In symptomatic cases, the presentations can vary from either abdominal pain, mass per abdomen, obstructive hydronephrosis and Meig's syndrome.[6,7]

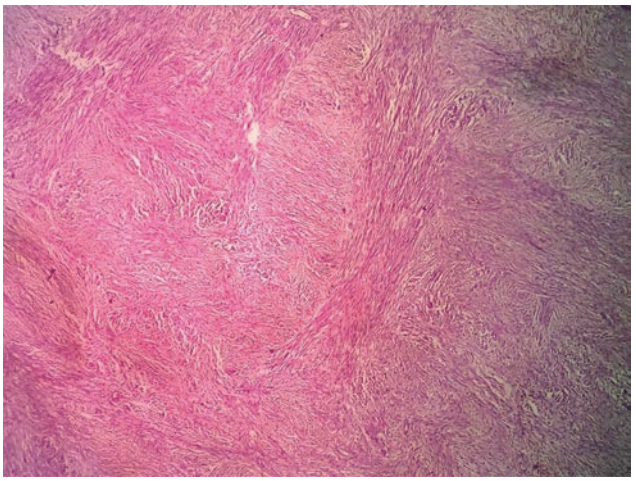

Fig. 3 Smooth muscle cells in interlacing bundles and fascicles, Hematoxilin & Eosin, 10x.

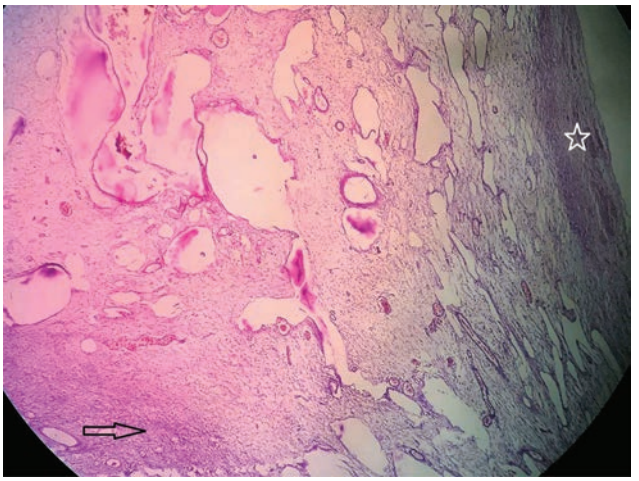

Fig. 5 Compressed ovarian stroma (star) with rete ovary and tumor below (arrow), Hematoxilin & Eosin, 10x.

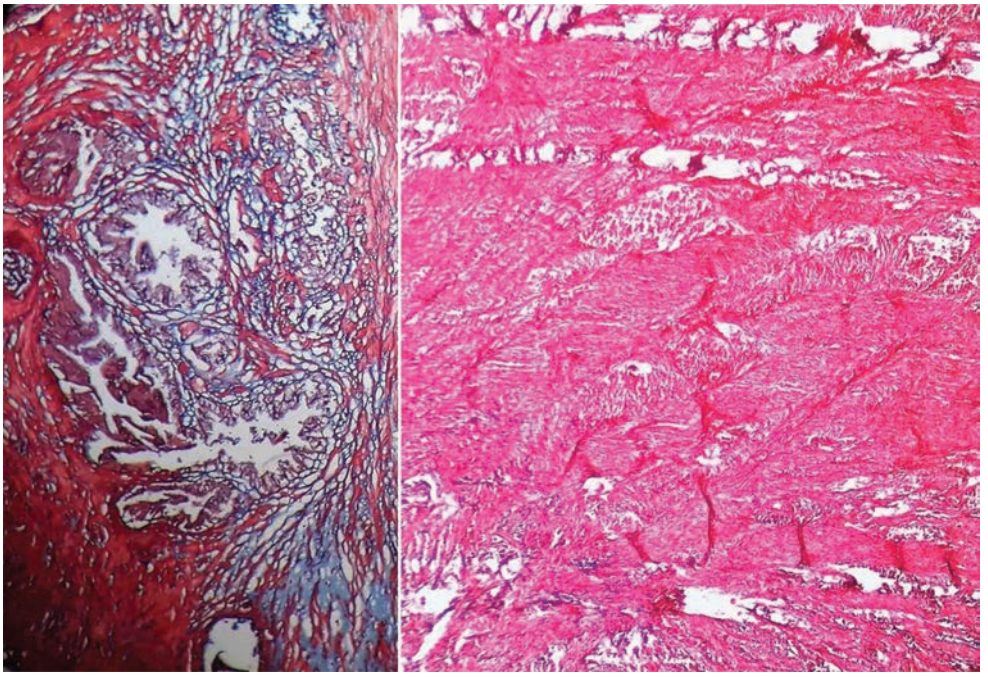

Fig. 4 Masson's trichrome stain—intense red positivity for smooth muscle cells (right), control (left).

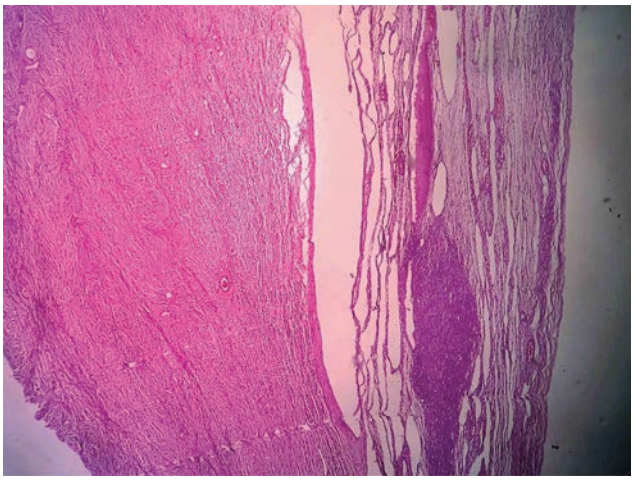

Fig. 6 Compressed ovarian tissue at the periphery of tumor, Hematoxilin & Eosin, 10x.

In our case study, patient was 39 years old and presented with abdominal pain and distension of 2-month duration.

Usually, primary ovarian leiomyoma are unilateral with an average of 3-cm diameter with no predilection for left or right ovary. A few bilateral cases have been reported, most of them occurred in patients between the ages of 16 and 25 years.[3] Unilateral ovarian leiomyomas are commonly associated with other ipsilateral or contralateral ovarian lesions. This association was found in 40% of the cases (mean age 45.8 years) reported by Doss et al. Furthermore, association with a concomitant uterine leiomyoma is also commonly seen, suggesting a common pathogenesis of the two tumors. It is most often diagnosed by chance during routine physical examination or incidentally at surgery or autopsy.[8] Radiological investigations which can aid in diagnosis include USG abdomen and pelvis and MRI.[3] MRI is potentially useful for the diagnosis of ovarian leiomyomas, which, like uterine leiomyomas, have intermediate signal intensity on T1 and low signal density on T2.[4]

In our case, ovarian leiomyoma was unilateral. USG abdomen and pelvis was done which showed a 12 ×12 cm, large heterogenous, hypoechoic, solid mass in the right adnexa with minimal vascularity. Periphery of the mass showed a cystic component, probably ovarian tissue. A differential diagnosis of right broad ligament fibroid and ovarian fibroma was given. Patient also had an associated intramural fibroid in the uterine corpus.

The common surgical approach of ovarian leiomyomas in middle-aged to elderly patients is by total abdominal hysterectomy with bilateral salpingo-oophorectomy. For symptomatic and large bulky masses, complete resection of the tumor is recommended. For bilateral ovarian leiomyomas, bilateraloophorectomy is often required. For patients who have not completed their family, ovariectomy of the affected ovary is done.

In our case, the patient had completed her family and hence total abdominal hysterectomy with bilateral salphingo-oophorectomy was done.[5,9]

The gross characteristics of ovarian leiomyomas are variable. Outer surface may show a nodular external surface with congested areas. Cut section shows areas with whorled appearance. Compressed ovarian tissue also may be seen within the capsule. While some of them are solid, others have cystic components or are predominantly cystic. Hemorrhage, calcification, and/or hyalinization may be seen.

Microscopically, ovarian leiomyoma is identical to its uterine counterpart and shows smooth muscle cells in interlacing bundles and fascicles composed of spindle-shaped cells with rod-shaped bland nuclei interspersed with foci of collagen deposition, with no evidence of any nuclear atypia or pleomorphism. Mitotic figures if present are very sparse.[1] The mass may show normal ovarian stroma at the periphery.

In our case, histopathological findings of right adnexal mass showed smooth muscle bundles arranged in interlacing bundles and fascicles suggesting leiomyoma. Compressed ovarian tissue showed ovarian stroma and hilar vessels at the periphery of the mass.

The main differential diagnosis includes ovarian fibroma, thecoma, broad ligament leiomyoma extending into the hilum of ovary, and pedunculated subserosal leiomyoma or wandering leiomyoma.[7] On Masson's trichrome stain (MTS), the tumor cells stain red which indicates the smooth muscle nature of the spindle cells.[10] In our case also, MTS showed smooth muscle nature of the tumor which ruled out fibroma of ovary. Broad ligament was normal and free from the mass intraoperatively and was not removed during surgery excluding extension of broad ligament leiomyoma into ovary. There was no evidence of subserosal fibroid on the surface of uterus; rather a tiny intramural fibroid was present, thus ruling out wandering leiomyoma.

Immunohistochemical study with smooth muscle specific staining (SMA and h-Caldesmon) can aid in diagnosis.[12] However IHC was not performed in our study as morphology and MTS was sufficient for diagnosis.

Primary ovarian leiomyoma has a good prognosis. Our patient is doing well since 2 months post surgery.

Conclusion

Leiomyoma is a benign tumor and its most common site of occurrence is the uterus. Primary ovarian leiomyoma is a very rare. It should be considered in the differential diagnosis when an ovarian tumor shows a solid, whorled appearance and microscopy showing spindle cells in fascicles. A careful search for the normal ovarian tissue should be made if the broad ligament is free. Despite its characteristic morphology, it needs to be distinguished from other benign tumors like ovarian fibroma, thecoma, broad ligament fibroid, leiomyoma extending into the hilum of ovary, and pedunculated or wandering leiomyoma. Histological examination is the gold standard for diagnosis. Masson's trichrome stain can aid in diagnosis.

Conflict of Interest
None declared.

References

1 Gilks B. Uterus corpus. In: Rosai and Ackerman, eds. Surgical Pathology. 11th ed. Philadelphia, PA: Mosby Elsevier; 2018: 1623–1688
2 Taskin MI, Ozturk E, Yildirim F, Ozdemir N, Inceboz U. Primary ovarian leiomyoma: a case report. Int J Surg Case Rep 2014;5(10):665–668
3 Choi SY, Park JS, Lee JW, Kim BG, Bae DS. A rare case of ovarian leiomyoma with atypical Meig's syndrome. Korean J Obstet Gynecol 2012;55(4):285–289
4 Fasih N, Prasad Shanbhogue AK, Macdonald DB, et al. Leiomyomas beyond the uterus: unusual locations, rare manifestations. Radiographics 2008;28(7):1931–1948
5 Manohar V, Pai VD, Kerudi SS, Ravindranath S, Tumbal S. Giant primary ovarian leiomyoma—a diagnostic dilemma. J Anesth Surg 2016;3(2):148–150
6 Kelekci S, Eris S, Demirel E, Aydogmus S, Ekinci N. Lipoleiomyoma of the uterus and primary ovarian leiomyoma in a postmenopausal woman: two rare entities in the same individual. Case Rep Pathol 2015;2015:564846
7 Rajabi P, Hani M, Bagheri M, Mirzadeh F. Large ovarian leiomyoma in young woman. Adv Biomed Res 2014;3:88
8 Abdessayed N, Salah M, Jouini R, Koubaa W, Brahim E, Debbiche AC. Primary ovarian leiomyoma in pregnant woman: an uncommon tumor with an unusual presentation. World J Pathol 2017;6:13–16
9 Kaya AE, Okten SB, Basbug A, Gamsizkan M, Dogan O, Baki H. Unilateral ovarian leiomyoma in a 14 year old adolescent: a rare case report. Arch Clin Exp Med 2018;3(1):38–40
10 Sheelaa WG, Kasinathan A. Primary ovarian leiomyoma in a postmenopausal woman: a case report. Int J Reprod Contracept Obstet Gynecol 2014;3(2):466–468