Case Report

Unilateral primary ovarian leiomyoma masqueraded as ovarian fibroma: A histopathological diagnosis

Sushma Bharti[1], Sudeep Khera[1], Charu Sharma[2], Arsha Balakrishnan[1]

Departments of [1]Pathology, and [2]Obstetrics and Gynecology, AIIMS, Jodhpur, Rajasthan, India

Abstract

Leiomyoma is the commonest benign mesenchymal tumor of the uterus, which can be developed at any site where smooth muscle cells are found. The broad ligament is the most common and ovary is one of the rarest extrauterine sites, accounting for 0.5–1% of all benign ovarian tumors. Herein, we report a case of ovarian leiomyoma in a perimenopausal female, clinically presented with heavy menstrual bleed, radiologically diagnosed as subserosal uterine fibroid. Intraoperatively, it was considered as ovarian fibroma but finally diagnosed as ovarian leiomyoma on histomorphology, which was confirmed on special stains and immunohistochemistry. A review of literature showed that less than 100 cases of primary ovarian leiomyomata have been reported until now. Ovarian leiomyoma is usually small, asymptomatic, and an incidental finding mostly, usually synchronously seen with uterine leiomyoma. Ovarian leiomyoma is a rare tumor, often misdiagnosed prior to surgical removal and it should be differentiated from other spindle cell neoplasm and solid tumors of the ovary.

Keywords: Extrauterine, fibroma, leiomyoma, ovary, perimenopausal

Introduction

Leiomyoma including its variants is the commonest benign mesenchymal tumor of the uterus.[1] It can be seen in the extrauterine sites. The broad ligament is the commonest extrauterine site[2] with an incidence rate of less than 1%.[3] Other unusual sites are round ligament, ovarian ligament, vulva, ovaries, urinary bladder, and urethra.[4] Ovarian leiomyoma although the commonest mesenchymal tumor of the ovary is one of the rarest benign tumors of the ovary.[5] To date, less than 100 cases have been reported and most of them were asymptomatic. Herein, we report a case of unilateral primary ovarian leiomyoma synchronous with uterine leiomyomata in a symptomatic premenopausal woman.

Address for correspondence: Dr. Sudeep Khera, Department of Pathology, AIIMS Jodhpur, Basni, Rajasthan - 342 005, India.
E-mail: drsudeepkhera@gmail.com

Received: 25-12-2020 **Accepted:** 23-05-2021
Published: 30-09-2021

Access this article online

Quick Response Code:

Website: www.jfmpc.com

DOI: 10.4103/jfmpc.jfmpc_2546_20

Case Report

A 42-year-old female was admitted for heavy menstrual bleeding and dysmenorrhea—clinically, considered as abnormal uterine bleeding. Endometrial biopsy showed hyperplastic endometrium with no evidence of malignancy. Complete blood count and other laboratory parameters and serum markers were normal. Transvaginal sonography showed a solid mass measuring 4 cm × 4 cm, abutting the uterus and left adnexa, suggesting subserosal fibroid. Intraoperatively, it was a 10-week size uterus with multiple subserosal fibroids and a solid mass along the left ovary, distinctly separated from the uterus with no adhesion or infiltration to the surrounding structures. The right ovary and bilateral fallopian tubes were normal. We received a bilateral salpingoophorectomy specimen. Uterus showed multiple subserosal and intramural fibroid. The Left ovary, measuring 7 cm × 7.5 cm × 4 cm, was totally replaced by nodular gray-white solid encapsulated homogenous mass showing a whorling pattern on cut surface. There was no evidence of necrosis and hemorrhage [Figure 1]. Sections from ovarian mass showed

How to cite this article: Bharti S, Khera S, Sharma C, Balakrishnan A. Unilateral primary ovarian leiomyoma masqueraded as ovarian fibroma: A histopathological diagnosis. J Family Med Prim Care 2021;10:3494-7.

typical leiomyoma histomorphology having intersecting short fascicles of spindle cells similar to the uterine leiomyoma. There was no evidence of increased mitosis, atypia, and necrosis, ruling out the possibility of leiomyosarcoma [Figures 2 and 3]. Because of the rarity of this tumor in the ovary, special stain and immunohistochemistry was done and the tumor was Masson trichome stain positive [Figure 4] and immunopositive for desmin, smooth muscle actin (SMA), and l-caldesmon [Figures 5–7], hence, confirming the diagnosis of ovarian leiomyoma [Figure 2] and ruling out the possibility of fibroma and other sex-cord stromal tumors and low-grade Gastrointestinal stromal tumor (GIST). The Endometrium showed features of hyperplasia with focal atypia. The myometrium showed extensive areas of adenomyosis and leiomyomata. The post-surgical period was uneventful.

Discussion

Primary ovarian leiomyoma is a rare benign tumor and accounts for 0.5–1% of all benign ovarian tumors,[5] which was first described in the year 1862 by Fallahzadeh *et al*.[6] A review of the literature shows that fewer than 100 cases of primary ovarian leiomyomata have been reported till 2020.[7] Most of them are usually small (<3 cm), and are asymptomatic, and therefore, detected incidentally either during routine physical examination, incidentally at surgery, or at autopsy after a histopathological examination. If the tumor is large, it may cause pelvic pain, palpable mass, or sometimes presents as acute abdomen due to torsion or necrosis in the tumor. In the present case, the female presented with a heavy menstrual bleed. Ovarian leiomyoma may be associated with Meigs' syndrome.[8] Serum levels of CA-125, CA 19-9, and Carcinoembryonic antigen (CEA) are within the normal limit. Ovarian leiomyoma synchronously seen with uterine leiomyoma in 78% of the cases suggests an identical hormonal stimulation.[9,10] The age group for both the ovarian and uterine leiomyoma is similar, having a very wide range, but only a few cases have been reported in postmenopausal women.[7] Both macroscopically and microscopically, ovarian leiomyoma resembles the uterine counterpart. Bilateral ovarian leiomyomya is seen in pediatric and young female and that are usually not associated with uterine leiomyoma counterpart as if seen in

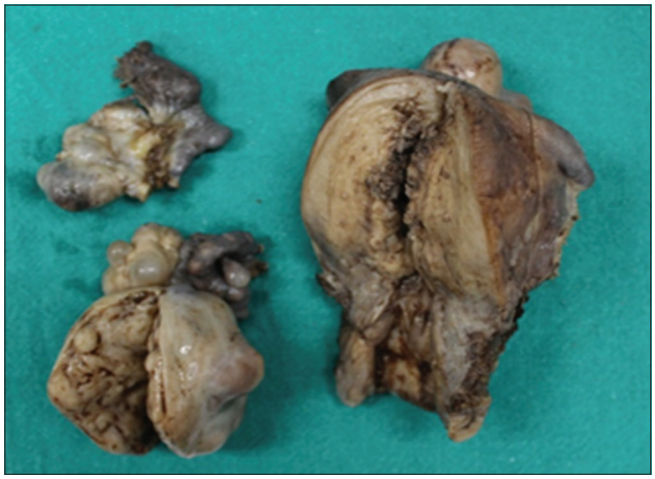

Figure 1: Gross image of the uterus and unilateral ovarian leiomyoma

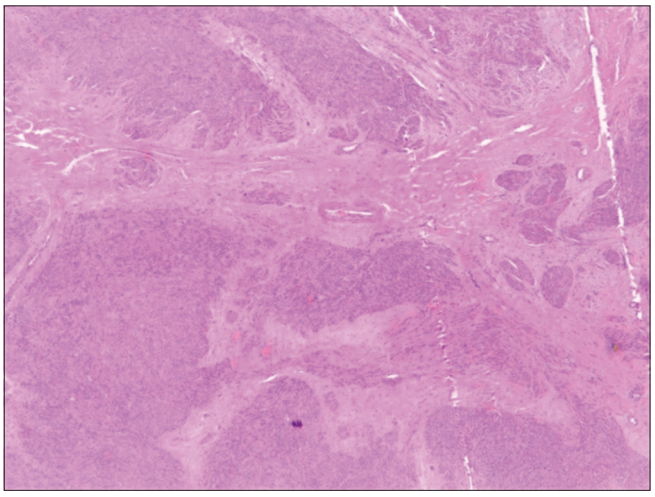

Figure 2: Histomorphology of ovarian leiomyoma (100X)

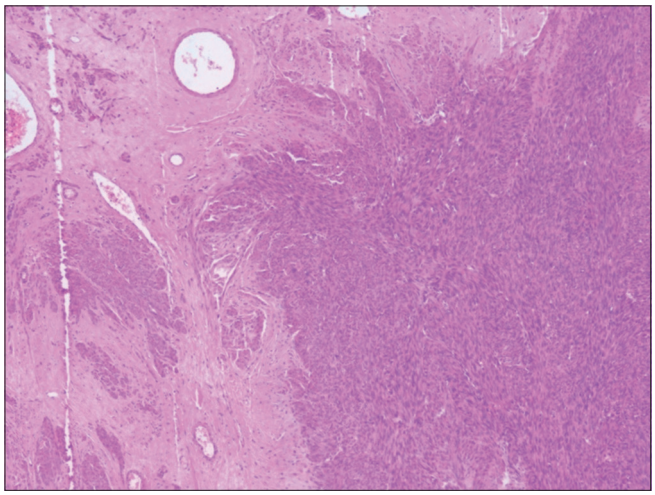

Figure 3: Histomorphology of ovarian leiomyoma (200X)

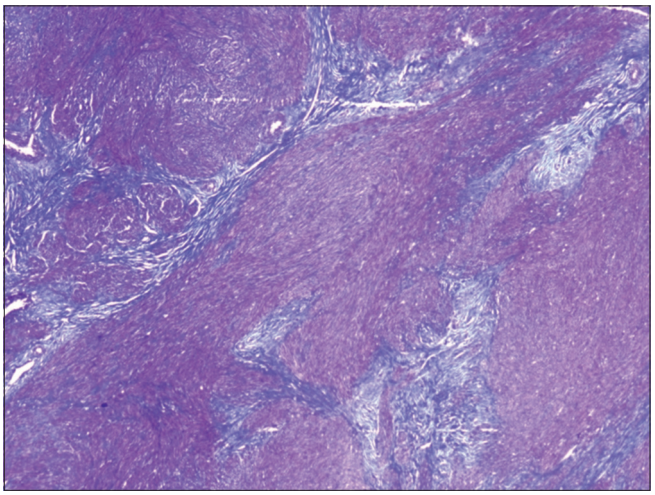

Figure 4: Masson trichrome stain

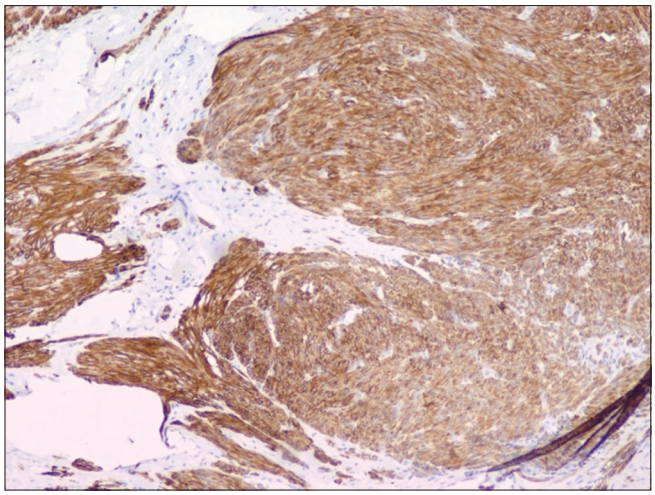

Figure 5: Desmin (200X)

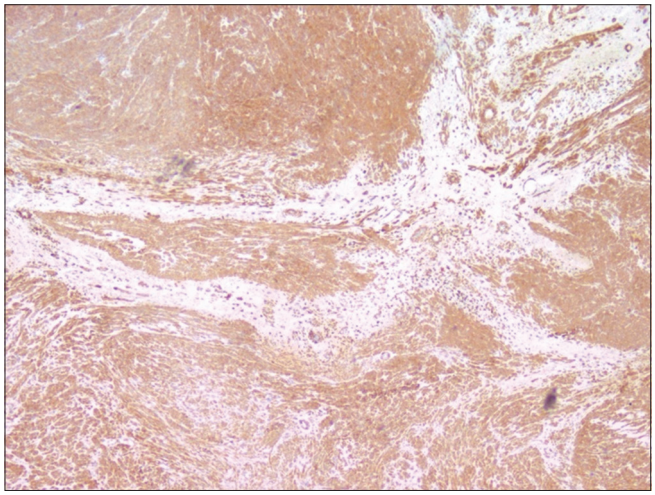

Figure 6: SMA (100X)

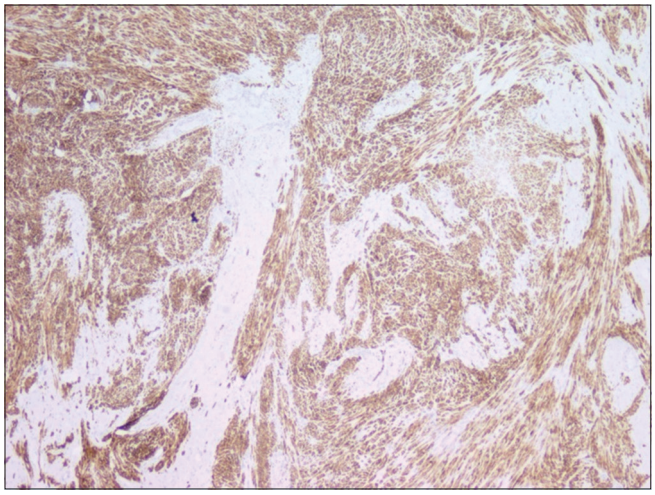

Figure 7: L-Caldesmon (100X)

perimenopausal female.[11] In the present case also there was unilateral ovarian leiomyoma with concomitant uterine leiomyoma presented with heavy menstrual bleed in a perimenopausal woman. A Few cases of ovarian leiomyoma in pregnant females are also reported in the literature. However, the exact cause of ovarian leiomyoma is unknown, but it is thought that they arise from the smooth muscle and include ovarian hilar blood vessels, ovarian ligament, smooth muscle cells or multipotential cells in the ovarian stroma, undifferentiated germ cells, cortical smooth muscle metaplasia, smooth muscle metaplasia of endometrial stroma, smooth muscle present in mature cystic teratomas, smooth muscle in the walls of mucinous cystic tumor and metastasizing uterine leiomyoma to the ovary.[12,13] There are no pathognomonic symptoms or characteristic imaging findings, and hence, it is a diagnostic challenge for the clinicians, and is mostly misdiagnosed. Therefore, it should be considered in the differential diagnosis of solid ovarian masses. Although, the treatment of ovarian leiomyoma is surgical resection only, the malignant ovarian tumors are commoner than ovarian leiomyoma in all age groups, including the reproductive age group, and are associated with a poorer prognosis. Therefore, the decision of an ovary preserving surgery should be taken carefully after a meticulous evaluation, considering the possibility of malignancy. An immunohistochemical analysis is recommended for definitive diagnosis. Leiomyoma is differentiated from fibroma-thecoma by Masson trichrome stain and desmin as SMA is positive in the fibrous tumor also. Thecoma is positive for lipid stain and inhibin. In the present study, leiomyoma was within the ovary and was completely encircled by the ovarian capsule, with no adhesion to the uterus or fallopian tube along with multiple intramural and subserosal uterine leiomyomas. Lerwill MF *et al.*[14] found 22 cases of ovarian leiomyoma out of which 12 cases were associated with uterine leiomyomas with histological features similar to that of their uterine counterparts, similar to this study while Agarwal *et al.*,[5] stated that primary ovarian leiomyoma has to be entirely within the ovary only with no similar lesions in the uterus or elsewhere. In the present case, ovarian mass was positive for Masson trichrome, desmin, SMA, and l-caldesmon [Figure 2], confirming the smooth muscle origin and ruling out the possibility of fibroma and thecoma.

Conclusion

Leiomyoma is a benign mesenchymal tumor that is frequently seen in the uterus but it rarely occurs in the ovary. The diagnosis of ovarian leiomyoma both preoperatively and intraoperatively is difficult because this condition is clinically indistinguishable from subserosal leiomyoma, ovarian fibroma, and other solid tumors of the ovary. Therefore, to assess a primary ovarian spindle cell tumor more accurately, histopathological examination along with immunohistochemical analysis and special stain is recommended.

Declaration of patient consent

Informed consent for patient information and images to be published was provided by the patients.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. International Agency for Research on Cancer. WHO Classification of Tumours of the Female Reproductive Organs (IARC WHO Classification of Tumours). 4th ed. Lyon: WHO/IARC Press; 2014.
2. Bhatta N. Tumours of the corpus uteri. Jeffcoats Principles of Gynaecology. 6th ed. London: Arnold Printers; 2001. p. 470.
3. Parker WH. Uterine myomas: An overview of development, clinical features, and management. Obstet Gynecol 2005;105:216-7.
4. Rajesh S, Adam M. Benign metastasizing leiomyoma: "A sheep in wolf's clothing". Community Oncol 2013;10:122-5.
5. Agarwal R, Kumar M, Agarwal L, Agarwal KK. A huge primary ovarian leiomyoma with degenerative changes: An unusual presentation. J Clin Diagn Res 2013;7:1152-4.
6. Fallahzadeh H, Dockerty MB, Lee RA. Leiomyoma of the ovary: Report of five cases and review of the literature. Am J Obstet Gynecol 1972;113:394-8.
7. Meel M, Hemrajani D, Kumar M, Agnani B. Symptomatic primary ovarian leiomyoma in a postmenopausal woman: A rare entity. J Midlife Health 2020;11:175-7.
8. Kurai M, Shiozawa T, Noguchi H, Konishi I. Leiomyoma of the ovary presenting with Meigs' syndrome. J Obstet Gynaecol Res 2005;31:257-62.
9. Lim SC, Jeon HJ. Bilateral primary ovarian leiomyoma in a young woman: Case report and literature review. Gynecol Oncol 2004;95:733-5.
10. Pandit MJ, Watson NR, Mackenzie IZ. Leiomyoma of the ovary. J Obstet Gynaecol 1997;17:503-4.
11. Wei C, Lilic N, Shorter N, Garrow E. Primary ovarian leiomyoma: A rare cause of ovarian tumor in adolescence. J Pediatr Adolesc Gynecol 2008;21:33-6.
12. Patne SC, Kumar M, Raghuvanshi S, Agarwal NR. Unilateral primary ovarian leiomyoma with degeneration masquerading as ovarian malignancy. World J Surg Res 2013;2:50-3.
13. Kataria SP, Chawla N, Singh G, Kum S. Ovarian leiomyoma associated with serous cystadenoma – A case report of an uncommon entity. Global J Pathol Microbiol 2014;14:38-42.
14. Lerwill MF, Sung R, Oliva E, Prat J, Young RH. Smooth muscle tumours of the ovary: A clinicopathologic study of 54 cases emphasizing prognostic criteria, histologic variants, and differential diagnosis. Am J Surg Pathol 2004;28:1436-51.