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Case Report

Primary ovarian leiomyoma: a diagnostic dilemma

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ABSTRACT

Primary ovarian leiomyoma is an uncommon entity accounting for 0.5-1% of all tumors and 4% of all the ovarian neoplasms. Ovarian leiomyoma is a benign, smooth muscle, solid tumors that occur in women aged 20-56 years. Because of its benign nature the prognosis for patient is good. A case of primary ovarian leiomyoma in a pre-menopausal female is to be presented here. A 49 years old female, married for 24 years came to gynaecology out-patient department with complaints of lower abdominal pain and abnormal uterine bleeding without dysmenorrhea and soakage of 4-5 pads per day since 1 month. On per abdomen examination non-tender firm to hard mass of 16 weeks gravid uterus size with regular margins, per speculum examination revealed cervicitis, per vaginal examination revealed the same mobile mass in left fornix and the uterus found to be normal size and to be deviated toward right side. CA-125 level found to be 10.71 and ultrasonically an ovarian mass of size $10 \times 8 \times 4$ cm was revealed. The decision of total abdominal hysterectomy with bilateral salpingo-oophorectomy was taken. The histopathology of ovarian mass confirmed to be the diagnosis of primary ovarian leiomyoma. This report highlights a case of primary ovarian leiomyoma in a pre-menopausal woman with a postoperative pathology report as a definitive diagnosis.

Keywords: Leiomyoma, Benign, Pre-menopausal, Total abdominal hysterectomy

INTRODUCTION

Primary ovarian fibroid is a very rare and benign mesenchymal tumor which is seen between 20 to 65 years of age. Ovarian leiomyoma has no side predilection and can be unilateral or bilateral, rather majority of bilateral cases occur in young patients. It is usually originated from the smooth muscles of hilar vessels, undifferentiated germ cells in ovarian stroma, stroma of endometriosis, ovarian ligament, and remnant of Wolffian body. These are mostly the asymptomatic tumor that occur in premenopausal women. If symptomatic, it may present with lower abdominal pain, ascites, pelvic mass, elevated CA-125, or Meigs syndrome.

Ultrasonography and MRI are preferred modalities, though many times it is difficult to differentiate it from other mesenchymal fibrous tumor such as fibroma or fibrothecoma.¹ Pre operative diagnosis of this tumor is

really difficult but post operative histopathological examination is gold standard for the diagnosis.⁷

Radical surgeries with Adnexectomy or total abdominal hysterectomy and salpingo-oophorectomy is usually carried out for ovarian leiomyoma.⁶

This is a case report of presentation of a benign tumor ovarian leiomyoma in a pre-menopausal woman.

CASE REPORT

A 49 years old female, married for 24 years came to an outpatient department of obstetrics and gynaecology of government medical college and hospital, Nagpur. Patient had complaints of lower abdominal pain and abnormal uterine bleeding without dysmenorrhea and soakage of 4-5 pads per day for 1 month. The patient's previous cycles were regular with normal flow. Obstetric history of the

patient includes 2 live births (both FTND/ uneventful), 2 died after birth and 2 abortions, none associated with the current presentation.

On per abdomen examination uterus measures up-to of 16 weeks gravid uterus size, with no local tenderness, Per speculum examination revealed cervicitis, per vaginal examination revealed the same mass of gravid uterus size 16 weeks, with firm to hard in consistency with regular margin non-tender felt in left fornix, uterus was of normal size and deviated toward right side.

Basic work up of the patient was done with CA-125 level found to be 10.71. Ultrasonography revealed a well-defined round to oval heterogeneously hypoechoeic lesion of size $10\times6.4\times7.7$ cm noted arising from fundus of the uterus, few calcific foci noted within, few necrotic areas seen within, not taking vascularity suggestive of subserosal uterine fibroid. There is no free fluid noted in the present scan and had no extension to omentum, liver, spleen, and intestine. Decision for laparotomy taken.

Intraoperatively, the uterine size was bulky with left ovary and Fallopian tube normal, there was a well circumscribed mass of around 15×10×5 cm arising from right ovary, with smooth surface and regular margins, normal ovarian tissue could not be identified, there was no free fluid in the abdomen, the surrounding viscera including the intestine, omentum, liver, spleen found to be normal. Decision for total abdominal hysterectomy with bilateral salpingo-oophorectomy was made.

On cut section, the mass was having fibers arranged in a whorled trabecular pattern, with no hemorrhages, necrosis or cystic spaces. The specimen was sent for histopathology examination. The histopathology shows predominantly presence of smooth muscle and spindle cells with no evidence of nuclear atypia, mitosis, or necrosis. These features were consistent with ovarian leiomyoma.

Post-operative period was uneventful. Patient was discharged after 5 days post-operatively and was asked to follow up. At the 6 month follow up, the patient had no complaints and no signs of tumor recurrence.



Figure 1: Specimen with left sided primary ovarian leiomyoma.



Figure 2: Cut section of primary ovarian leiomyoma.

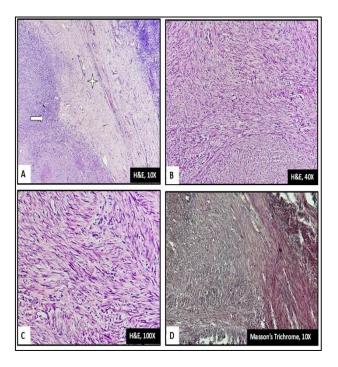


Figure 3 (A-D): Histopathology of primary ovarian leiomyoma.

DISCUSSION

Ovarian leiomyoma accounts for 0.5%-1% of all benign tumors.⁵ This is usually seen between 20 to 65 years of age and is detected incidentally.⁶ The majority of these tumors, 85% is discovered in perimenopausal and postmenopausal women together. Rather in menopausal women the incidence accounts just for only 16%.¹ About 78% of ovarian leiomyoma were associated with uterine leiomyoma and both may have a common trigger.⁷ Many of the studies has shown that fewer than 100 cases of primary ovarian leiomyomata have been reported till 2020.⁹ Ovarian leiomyoma is usually originated from smooth muscles of hilar vessels, undifferentiated germ cells in ovarian stroma, ovarian ligament, stroma of endometriosis, and remnant of Wolffian body.³ The level

of estrogen in blood promotes the growth of ovarian leiomyoma because as the level of estrogen decreases after menopause so is the size of tumor concomitantly. 4,2

Primary ovarian leiomyoma may often be misdiagnosed preoperatively as pedunculated uterine myoma, ovarian fibroma, or even ovarian endometrioma.⁸

Mostly the ovarian leiomyoma is usually asymptomatic but 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. Some of the patients may also present with Meigs syndrome with hydro thorax and ascites. ⁵ The tumor markers such as CA-125, CA19-9, and CEA (Carcinoembryonic antigen) are mostly within normal limits whereas in some cases such as the giant leiomyoma may present with increase in the level of CA-125.9,1 Ultrasonography and magnetic resonance imaging (MRI) cannot be used as a modality of choice to differentiate between ovarian tumors.7 The Ultrasound report may depict the tumor as solid tumor, such as mass or a solid mass with cystic component, which is nonspecific and can mimic neoplasia. 10 Primarily these lesions may appear solid on MRI fields.1 Leiomyoma on MRI shows low intensity on T1 and T2 weighted images and early enhancement on contrast MRI, on the other hand fibroma or thecoma shows poor enhancement on contrast MRI.5

Differential diagnosis such as fibroma, fibrothecoma, primary leiomyoma, thecoma, cystadenofibroma, Brenner's tumor, and sclerosing tumor which all produces hypo-intense images on MRI should also be kept in mind.¹

The gold standard method to confirm the diagnosis is histopathology and immunochemical analysis.³ Haemotoxilin and Eosin stains are used to diagnose leiomyoma in view of the age of patient and location whereas IHC markers are used to rule out other differentials.⁸ Leiomyoma on IHC shows strong positivity with alpha smooth muscle actin (alpha SMA) and diffuse positivity with Desmin, whereas fibroma is negative for Desmin and positive for SMA, lastly Thecoma does not express SMA instead express Alpha inhibin and calretinin.⁶⁻⁸

The best treatment modality for ovarian leiomyoma is surgery with overdue consideration of patients age, tumor size, fertility needs, contra-lateral ovary and other factors. ¹⁰ In this particular case, total abdominal hysterectomy with bilateral Salpingo-oophorectomy was performed as the patient is perimenopausal and has already completed her family. Enucleation that is ovarian preserving technique is usually preferred in young patient. ¹⁰

CONCLUSION

Primary ovarian leiomyoma are rare tumors of unresolved origin and should be considered in the differential diagnosis of ovarian spindle cell tumors. The diagnosis of preoperatively ovarian leiomyoma both intraoperatively is difficult, therefore to assess a primary ovarian spindle cell accurately tumor more histopathological examination along with immunohistochemical analysis and special stain is recommended.

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