Case Report

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OVARIAN LEIOMYOMA- RARE TUMOR WITH UNUSUAL PRESENTATIONS: REPORT OF 2 CASES WITH BRIEF REVIEW OF LITERATURE

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ABSTRACT

Ovarian leiomyoma is a rare and incidentally detected neoplasm. It is usually reported in women of childbearing age. Clinically most patients are asymptomatic. Histopathologic examination is required to distinguish it from subserous leiomyomas and fibroma-thecoma group of ovarian tumors. We present 2 cases of ovarian leiomyoma along with brief review of literature. The first case of a 25 year old woman who presented with primary infertility and history of irregular menstruation and the second case of a 54 year old post menopausal lady with complaints of abdominal pain who was referred with the clinical impression of ovarian malignancy.

Key words: Ovarian leiomyoma, infertility, fibroma

INTRODUCTION

Ovarian leiomyoma was first described in the year 1862[1]. It is a rare benign tumor that accounts for 0.5 to 1% of all benign ovarian tumors[2]. Origin of ovarian leiomyomas is not yet established with many possibilities described in the literature. It is found to be frequently associated with uterine leiomyomas[3].

CASE REPORT

Case I: A 25 yrs old female, came with the complaints of primary infertility. She was married for four years, gave history of regular sexual intercourse with no usage of any contraceptives. She also had history of irregular menstrual cycles. General physical examination was normal. Per vaginum left adnexal mass was noted. Transvaginal sonography showed left tubo-ovarian mass and pelvic inflammatory disease, with clinical suspicion of chocolate cyst. On diagnostic laparoscopy, left ovary was enlarged with lobulations and glistening surface. There were no adhesions. It was not possible to preserve a part of affected ovary as no healthy parenchyma could be identified. Left fallopian tube, right ovary and tube were normal. Left salpingo-oopherectomy was done and specimen was sent for histopathology. Grossly, multiple nodular grey white tissue bits together weighing 76 gms, largest tissue bit measured 7x 3x2.5 cms. Cut section showed grey white areas with whorling pattern along with degenerative changes. No normal ovarian parenchyma was noted (Figure 1).

Microscopy showed a vague nodular neoplasm composed of intersecting fascicles of spindle shaped cells resembling smooth muscles, with oval and vesicular nuclei showing fine chromatin interspersed with collagen, blood vessels, stromal hyalinization and calcific deposits. No significant mitotic activity/ cellular pleomorphism/ necrosis noted. Ovarian parenchyma was preserved only at the periphery (Figure 2, 3, 4).

On Masson's trichrome stain fascicles of spindle shaped cells stained red in color confirming the muscle origin (Figure 5).

Fallopian tube showed normal morphology. Based on histopathology and special stain diagnosis of ovarian leiomyoma was given.

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Case II: A 54 year old female came with the complains of dull pain and distension in the lower abdomen for the last 4 years. There was no history of any discharge per vaginum and bowel/ bladder abnormalities. Work up done outside the hospital was suggestive of ovarian malignancy. Patient was referred to our hospital for further management. On examination, per abdomen a mass was felt in the subrapubic area with restrictive mobility. Per speculum, senile changes were noted. On per vaginum examination a mass was felt measuring approximately 10 cms across on the right side with restricted mobility. All hematological and biochemical investigations were within normal limits. Serum beta HCG, alpha fetoprotein, C.A 19.9, C.A 125 and carcinoembryonic antigen were normal. Transvaginal ultrasound revealed a solid pelvic mass measuring 11x9.2 cms with no doppler uptake. Ovaries were not visualized separately. On CT scan abdomen a well defined heterogeneously enhancing solid abdomino-pelvic lesion arising from the right adnexal region was seen with the possibility of ovarian malignancy. Patient underwent staging laparotomy, right ovariectomy, omentectomy and pelvic lymphadenectomy. On gross examination, specimen of ovary was weighing 365 gms and measuring 11x10x7.5 cms. Cut section showed a well circumscribed solid grey white mass measuring 7x7 cms with preserved ovarian parenchyma at the periphery (Figure 6).

Histology showed tumor composed of fascicles of spindle shaped cells with plump nuclei and few with moderate pale cytoplasm, arranged in sheets and storiform pattern with interspersed foci of collagen deposition, areas of hyalinization and cystic degeneration. No significant mitotic activity/ cellular pleomorphism/ necrosis noted (Figure 7, 8). Masson's trichrome stain confirmed the muscle origin (Figure 9).

Immunohistochemistry done showed tumor cells positive for smooth muscle actin. Ki-67 index was less than 1% (Figure 10, 11).

All the lymphnodes showed reactive changes. Based on histopathology, special stain and immunohistochemistry a diagnosis of ovarian leiomyoma was given.

DISCUSSION

Ovarian leiomyoma is a rare benign tumor accounting for 0.5 to 1% of all benign ovarian tumors[2]. About 80 cases have been reported in the literature worldwide till date[4]. It generally occurs in premenopausal women aged between 20 to 65 years[2,3]. Only about 16% of cases are reported to occur after menopause[5]. These tumors are usually unilateral, although a single case of bilateral

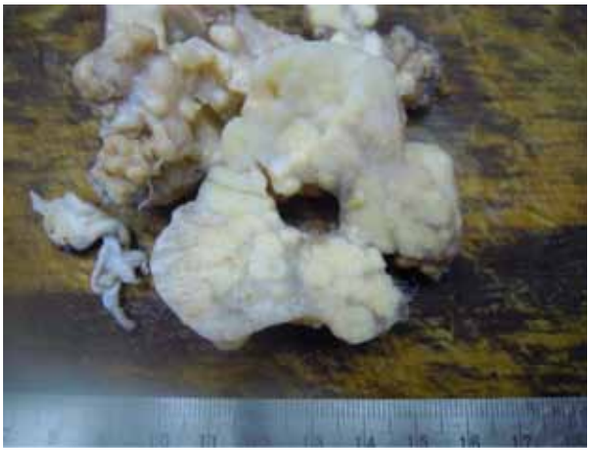

Figure 1: Well circumscribed ovarian tumor, grey white in color. Cut surface shows whorling pattern

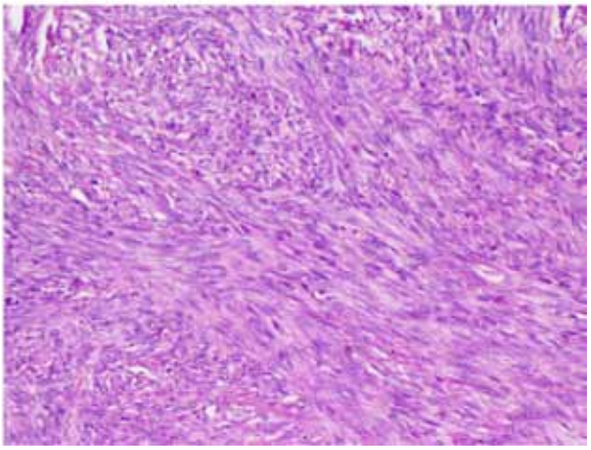

Figure 2: Tumor composed of intersecting bundles of spindle shaped cells (H&E, x100)

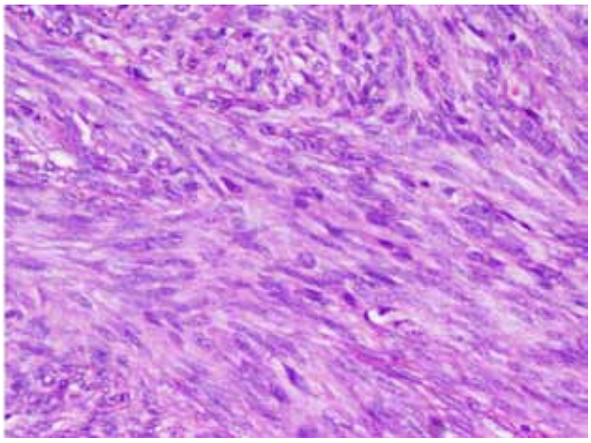

Figure 3: Spindle cells with oval and vesicular nuclei, fine chromatin, with no mitosis/ pleomorphism/ necrosis (H&E, x200)

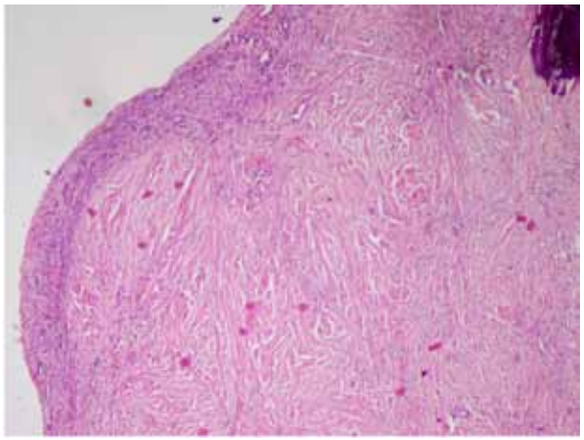

Figure 4: Preserved normal ovarian parenchyma at periphery, degenerative changes- stromal hyalinization and calcific deposits in the tumor (H&E, x40)

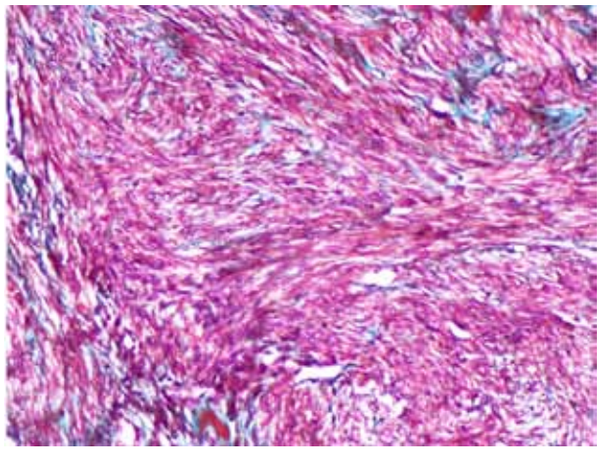

Figure 5: Fascicles of smooth muscle bundles stained red (Masson Trichrome, x100)

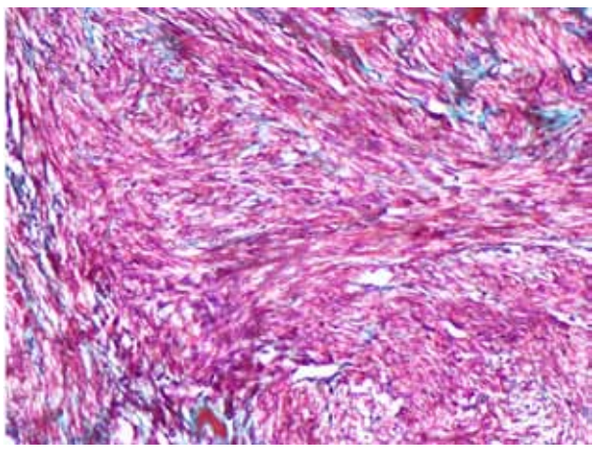

Figure 6: Well circumscribed solid grey white tumor with preserved ovarian parenchyma at the periphery

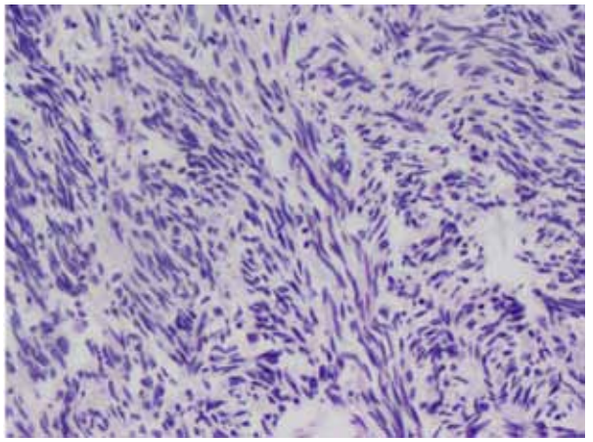

Figure 7: Fascicles of spindle shaped cells with plump nuclei and moderate pale cytoplasm (H&E, x 100)

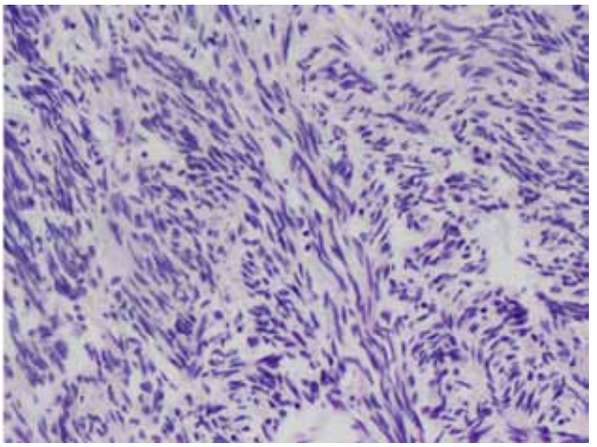

Figure 8: Fascicles of spindle shaped cells with no significant mitosis/ nuclear pleomorphism/ necrosis (H&E, x 200)

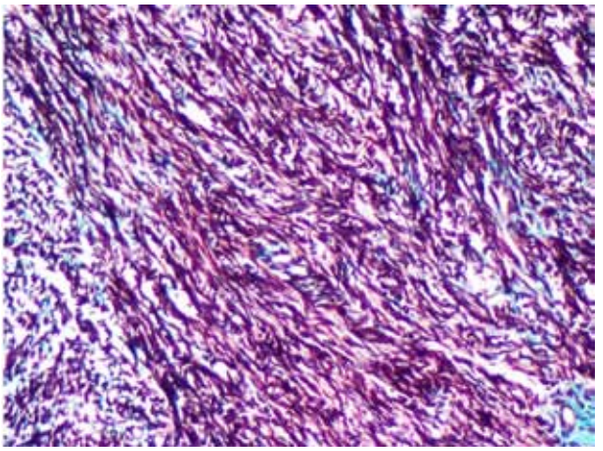

Figure 9: Tumor cells stained red confirming muscle origin (Masson Trichrome, x100)

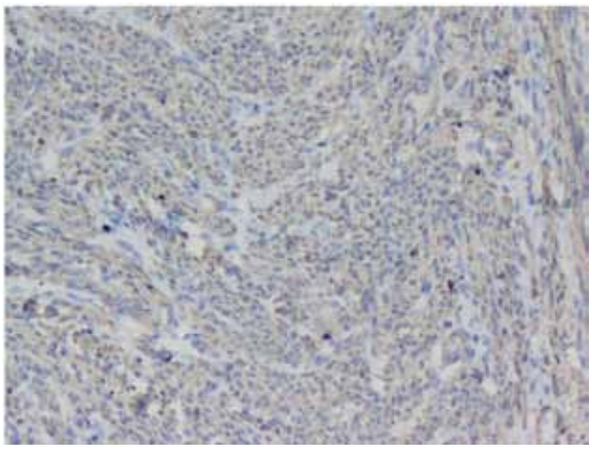

Figure 10: Diffuse Smooth Muscle Actin(SMA) positivity in tumor cells (x200)

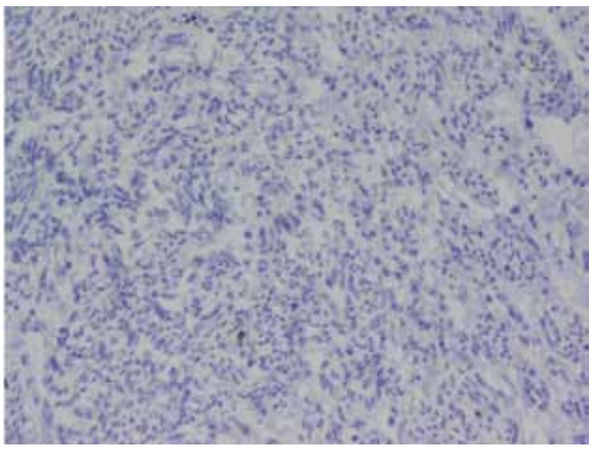

Figure 11: Ki-67 index less than 1% (x200)

ovarian leiomyoma in a 21-year-old woman has been documented in literature[6]. Bilateral involvement is uncommon in women over 35 years of age[2].

Origin of ovarian leiomyomas is not well established yet. The probable origin could be smooth muscle cells in the ovarian hilar blood vessels, cells in the ovarian ligament, smooth muscle cells or multipotential cells in the ovarian stroma, undifferentiated germ cells, or cortical smooth muscle metaplasia[2]. And the described probable origin from smooth muscle metaplasia of endometriotic stroma, smooth muscle present in mature cystic teratomas, and smooth muscle in the walls of mucinous cystic tumor may also explain their concomitant occurrence in certain ovarian lesions[2]. Uterine leiomyoma directly metastasizing to the ovary is another possible origin[7]. Our first case was not associated with uterine leiomyoma. Hence this was a primary lesion and not a parasitized or a metastasized uterine neoplasm. However, in second case since the cause of hysterectomy was not known, hence the possibility of associated uterine leiomyoma cannot be ruled out.

Most ovarian leiomyomas are asymptomatic and are found either during routine physical examination, incidentally at surgery or at autopsy. In symptomatic cases, clinical presentations like abdominal pain, palpable mass, hydronephrosis, hydrothorax, ascites and elevated CA-125 have been described[2,3]. The largest tumor, measuring 36x37x11cms and weighing 6855gms was reported in a 72-year-old nulliparous woman who presented with ascites and polymyositis[8]. In our first case, patient presented with primary infertility. Ovarian leiomyomas are described as a potential cause of compromised fertility[9], but further studies are needed for substantiation. Second case presentation with abdominal pain and palpable mass mimicking ovarian malignancy has also been described and reported in literature[5].

Ovarian leiomyomas are usually associated with uterine leiomyoma, probably suggesting an identical causative hormonal stimulant. They are identical to their uterine counterpart both grossly and on microscopy[7]. Degenerative changes such as hyalinization, calcification, and cyst formation may be seen in ovarian leiomyomas[3] as well and were noted in our cases. They are also described to be associated with certain other ovarian lesions like ovarian endometriosis and mucinous cystadenoma[2,10-12].

The correct diagnosis of ovarian leiomyomas requires recognition of its smooth-muscle origin. Differential diagnosis include tumors in the fibroma-thecoma group, tumors arising in the broad ligament and extending into the hilum of the ovary, myometrial fibromyomas becoming parasitic on the ovary and leiomyosarcomas. Ideally primary ovarian leiomyomas should be entirely within the ovary, with no similar lesions in the uterus or elsewhere[1,3]. Masson's trichrome stain can be used to distinguish smooth muscle and fibrous components of the tumor[7]. Immunohistochemistry (IHC) is confirmatory and used in doubtful cases as the cytoplasm of the spindle tumor cells stain positive for desmin and smooth muscle actin. Desmin shows diffuse positivity in leiomyomas, whereas fibromatous tumors are typically negative or only focally positive. Since smooth muscle actin is usually positive in both, hence not helpful in distinguishing the two. Thecoma tumor cells do not express smooth muscle actin and thus may be differentiated from leiomyomas which are strongly positive for this stain[2,7]. Primary leiomyosarcoma of the ovary is hypercellular with high mitotic activity (>10 mitotic figures per 10 high power fields), hence can be differentiated from its benign counterpart[3]. Our cases showed fascicles of smooth muscle cells which was confirmed on Masson's trichrome stain. Immunohistochemistry was done for the second case also confirmed the diagnosis.

Treatment of ovarian leiomyoma is surgical removal. These tumors are benign, hence least radical surgery should be preferred[1,7]. Conservative surgery with preservation of ovarian function and anatomy is considered to be the first treatment modality especially in patients of reproductive age[4].

CONCLUSION

Ovarian leiomyoma is a rare benign tumor with distinct histopathologic and immunohistochemical features. In our first case patient presented with primary infertility and the absence of a uterine counterpart of the lesion makes it primary ovarian origin. The origin of ovarian leiomyoma is still unresolved. Despite its rarity, it should be considered as a possible cause of infertility. Our second case mimicked ovarian malignancy in its presentation. Awareness of this entity can avoid unnecessary aggressive surgery. Immunohistochemistry may be required in its differentiation from other ovarian spindle cell tumors. Since, most of these tumors appear at reproductive age, ovary- preserving management is preferred.

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