



Case Report

Stromal luteoma in a case of multiple leiomyomatosis – A rare case report

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ABSTRACT

Stromal luteoma of the ovary occurs mostly in postmenopausal females and is very rare. They account for 60% Abnormal vaginal bleeding is the most frequent presentation, but endocrine symptoms and sometimes virilizing signs may also be observed. This tumour is a close mimicker of Leydig cell tumour. However, the stromal location and the absence of Reinke's crystalloids helps to confirm the diagnosis. This tumour is surrounded by an ovarian stroma, entirely composed of luteinized cells devoid of crystals of Reinke. We report an unusual case of a 41 year old female who presented with abdominal pain and menorrhagia. A TAH + BSO was performed and microscopy revealed a stromal luteoma of the right ovary with multiple intramural and subserosal fibroids. Preoperative diagnosis was a leiomyoma. However, microscopy revealed a stromal luteoma in the ovary. One third of the stromal luteomas are incidental findings. The preoperative and intraoperative diagnosis of this condition is challenging. Awareness about this condition among pathologists and clinicians may help in preventing misdiagnosis and overtreatment.

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1. Introduction

Sex-cord ovarian tumours are a rare kind of ovarian neoplasm. Steroid cell tumours that account for 0.1% of all primary ovarian tumours are also a sub-group of sex-cord ovarian tumours. Steroid cell tumours are classified into three groups, according to the origin of the cells that generate the tumour: stromal luteomas; Leydig cell tumours; and steroid cell tumours not otherwise specified (NOS). Stromal luteomas constitute 20 to 25%, Leydig cell tumours 20 to 25% and steroid cell tumours NOS constitute 50 to 60% of all steroid cell tumours.¹

Stromal luteoma of the ovary occurs mostly in postmenopausal females and is very rare. Abnormal vaginal bleeding is the most frequent presentation, but endocrine symptoms and sometimes virilizing signs may also be

observed.²This tumour is surrounded by an ovarian stroma, entirely composed of luteinized cells devoid of crystals of Reinke.

Steroid cell tumours can be seen in all ages, however the incidence increases in the fifth and sixth decades.³

Symptoms may differ depending on the hormones secreted from the tumour cell. Stromal luteomas are usually present with hyper-oestrogenic symptoms when Leydig cell tumours are mostly present with hyper-androgenism, and the NOS group is often seen with androgenic effects. However in the NOS group, hyper-oestrogenism may rarely be seen, as 25% of NOS tumours may have no hormonal activity.⁴

2. Case Report

A 41 years old female patient presented to the department of obstetrics and gynaecology with chief complaints of abdominal pain and menorrhagia. On further clinical

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and radiological examination she was diagnosed with large intramural posterior wall fibroid along with few sub centimetric intramural fibroids and a subserosal fibroid. Patient underwent a total abdominal hysterectomy and bilateral salpingo- oophorectomy [TAH&BSO]. We received the specimen and it was processed for histopathological examination and following findings were noted.

2.1. Macroscopically

The cut surface showed a big intramural fibroid measuring 6 x 5 x 4 cm along with few sub centimetric intramural and a subserosal fibroid. It was grey white in colour with a whorled appearance. Right ovary measuring 2.5x2x0.8cm. Cut surface showed a single cyst measuring 0.3x0.3cm. Also seen were an yellowish area measuring about 1x1x0.5 cm. Left ovary measuring 3x2x1cm. Cut surface show single cyst lumen. Bilateral tubes were unremarkable.

On Microscopic examination, the cervix showed features of chronic cervicitis, the endometrium showed features of a simple hyperplasia without atypia and the myometrium revealed multiple leiomyomatosis and a single subserosal leiomyoma.

On examination, the right ovarian stroma was replaced by sheets and nests of cells with individual cells showing round central nuclei with abundant eosinophilic cytoplasm with few showing a clear vacuolated cytoplasm. No Reinke crystalloids were seen. No mitosis/necrosis. The surrounding thin rim of ovarian parenchyma is unremarkable.

The left ovary showed a simple serous cyst. We gave a final impression of Multiple leiomyomatosis with Stromal luteoma of right ovary.

3. Discussion

Stromal luteomas are almost always less than 3 cm in diameter and, with rare exceptions, are unilateral. They are well circumscribed, solid, and usually grey-white or yellow, but one third of them have red or brown areas or are uniformly so.^{5,6}

Microscopic examination of a stromal luteoma reveals a more or less rounded nodule of cells of lutein type that generally contain relatively little lipid. Intracytoplasmic lipochrome pigment may be conspicuous. The nuclei are small and round with a single prominent nucleolus. Mitoses generally are rare. The cells may be arranged diffusely or in small nests or cords and are more or less completely surrounded by ovarian stroma.^{5,6} The diagnosis of stromal luteoma is supported in approximately 90% of the cases by the finding of stromal hyperthecosis elsewhere in the same or contralateral ovary which helps to differentiate it from steroid cell tumour not otherwise specific.



Fig. 1: Ovary- gross

One confusing feature, seen in about 20% of the cases, is focal degeneration, with the formation of irregular spaces that may simulate glands or vessels. These spaces may contain, or be surrounded by, lipid-laden cells and chronic inflammatory cells and may be associated with fibrosis. In some cases they contain red blood cells.⁷

The commonest differential is a Leydig cell tumour, but the stromal location of the tumour and the absence of Reinke crystalloids helps in the diagnosis of a stromal luteoma. The other differential is a theca lutein cyst. Theca lutein cysts always present as a follicle in the stroma, but this is not observed in stromal luteomas. Malignant melanomas can rarely mimic stromal luteomas and can be differentiated by S100 and human melanoma black-45 immunoreactivity in melanomas.⁷

Young (2011) showed that stromal luteomas occur mostly (80%) in postmenopausal women, and 60% of the patients present with hyper-oestrogenic abnormal bleeding. Only 12% of the patients with stromal luteoma have androgenic symptoms.^{3,8}

Young (2011) reported that stromal luteomas and Leydig cell tumours are mostly seen in postmenopausal women, whereas NOS are mostly seen in young premenopausal women (mean age 43). In our case it the age was 41 and the diagnosis was made in premenopausal women. Young et al, in their study, showed preoperative TV-US generally missed the diagnosis of a stromal luteoma and it was picked up a routine histopathological examination. Mean tumour diameters are 1.3cm for stromal luteomas, 2.4cm for Leydig

cell tumours and 8.4cm for NOS. According to the above study, stromal luteomas had the smallest diameter among the steroid cell tumours which was similar to our case which showed a diameter of 1cm.^{3,8}

Ovaries should be examined carefully with transvaginal ultrasonography to reveal any accompanying ovarian pathology in women with postmenopausal bleeding, and stromal luteomas should be considered as a cause for the postmenopausal bleeding, even though they are rare. Nevertheless, the transvaginal ultrasound is the most sensitive method for the detection of an ovarian tumour.⁹

In our case, stromal luteoma was an incidental finding which was missed on clinical and radiological examination. So, It is imperative for the clinician and the radiologist to be aware of this rare condition and to consider this as a possibility for heavy menstrual bleeding. This case report highlights the importance of a thorough pathological examination in the diagnosis of this condition which was missed both clinically and radiologically.

3.1. Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/ her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

4. Conflict of Interest

The authors declare that there is no conflict of interest.

5. Source of Funding

None.

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