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Case Report

Low grade endometrial stromal sarcoma: A case report

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ABSTRACT

Endometrial stromal sarcoma (ESS) is an uncommon and confusing malignant tumour amongst all uterine sarcomas occurring in the peri-menopausal age group. We report a case of low-grade ESS in a 47-year-old woman, presented with lower abdominal pain related with lower abdominal mass. Earlier USG report exposed multiple intramural fibroids. Since preoperative decision could not be determined, total abdominal hysterectomy with bilateral salpingoopherectomy was done. Histopathology examination of uterus revealed low grade endometrial stromal sarcoma. Post operatively PET CT was done which came out to be negative for metastasis. Patient is in regular follow up since then.

1. Introduction

Sarcoma is a rare malignant tumour of uterus, with an incidence of 1-2 cases per 100,000 establishing around 0.2% of uterine malignancies and nearly 20% of uterine sarcomas.[1] Based on WHO classification 2014, ESS can be classified in to four types such as endometrial stromal nodule, low-grade endometrial stromal sarcoma (ESS), high-grade endometrial stromal sarcoma and undifferentiated uterine sarcoma.[2] Majority of cases (90%) generally have abnormal uterine bleeding and enlargement of uterus (70%) as presenting complaints. Leiomyoma, adenomyosis or intrauterine polyps are the often misdiagnosed benign conditions through ultra-sonogram.[3] Generally diagnosis is established by histopathological assessment of tumour specimens post-operatively.[4] Hysterectomy has been the undisputed treatment of choice considering the post-operative finding and paucity of this condition.[5] Prognosis is generally poor but in early diagnosis prognosis is very good.

2. Case Presentation

A 47 year old female, came to out-patient department of Obstetrics and Gynecology with presenting complaints of lower abdominal pain and mass in lower abdomen for 2 months. She had lower abdominal pain for the past 2 months, a dull aching type of pain which was insidious in onset, intermittent in nature, aggravated on activity and relieved on taking medication with no radiating features. She has regular menstrual cycles and gives a history of Uterine fibroid of small size 10 years back but was not on follow up since then. Thorough clinical examination and investigations were done. On general examination, she was not pale, no pedal edema and no lymphadenopathy. Per abdominal examination was soft and no mass was palpable. Per speculum examination shows minimal erosions were present on the cervix and vagina was healthy. Whereas Per vaginal examination shows uterus anteverted and corresponding to 12 weeks size, bilateral fornices free and non tender.

Ultrasound Abdomen and Pelvis revealed multiple uterine fibroids, a calcified intramural fibroid with an

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ill defined margin and heterogenous echogenicity of size 3.1*3.2 cm in anterior myometrium. Another fairly defined heterogeneously hyper echoic polypoid mass with nodular myometrial extension of size 6.9*8.5 cm was noted in the left lateral wall/ posterior myometrium with diffuse myometrial thickening. After obtaining cardiology and anaesthesia fitness, patient was then planned for fractional curettage with cervical biopsy.(Figure 3)

Histopathology examination of cervical biopsy exposed fragments of tissue with stroma showing scattered inflammatory cells admixed with few foamy histiocytes. As certain judgment could not be determined, after discussing with the patient and her attenders, total abdominal hysterectomy along with bilateral salpingoophorectomy was done. Intraoperative findings revealed posterolateral fibroid of size 9x7 cm with cystic degeneration towards left side. It was an intracavitary mass which was thin and membranous with no solid components. Cut section of specimen showed internal haemorrhage and necrosis. Histopathology examination of uterus showed irregular cellular islands, forming permeative tongue like pattern of myometrial invasion with frequent vascular invasion, tumour cells are oval to spindle shaped with minimal cytologic atypia, vesicular chromatin and scant cytoplasm suggestive of low grade endometrial stromal sarcoma but regional lymph nodes could not be assessed. (Figures 1 and 2)

Postoperatively, CECT Abdomen and HRCT Thorax were done. CECT Abdomen showed post hysterectomy with bilateral sapling oophorectomy status, present study shows minimal collection in pelvis closely abutting the rectum and sigmoid colon. HRCT Thorax showed multiple randomly distributed soft tissue dense nodules in bilateral lungs - metastasis, fibroatelectasis with cystic bronchiectatic changes in apiece posterior segment of left upper lobe.

PET CT was done according to Surgical oncology opinion and the results were negative for metastasis. Patient is in regular follow up since then.

3. Discussion

Endometrial stromal sarcoma is an uncommon condition which affects about 0.2% of all reproductive tract malignancies. The current incidence is about 2 per million women whereas; the incidence rate is 700 per million women in case of endometrial cancer.[5]

Sarcoma of uterus generally affects postmenopausal women and low grade ESS occurs between 45 and 55 years. According to previous researchers, the presenting symptoms are comparable to uterine leiomyoma; abnormal vaginal bleeding, pelvic mass or abdominal pain can be present and some patients may be asymptomatic.[6] Majority of the cases will have abnormal uterine bleeding (90%), enlarged uterus (70%) and metastases (30%-50%).[7] In this index case, abnormal uterine bleeding was absent which is very

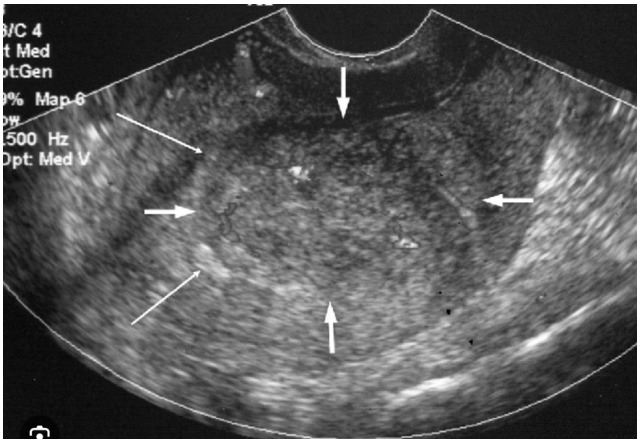

Figure 1: Ultrasound findings of low grade endometrial stromal sarcoma

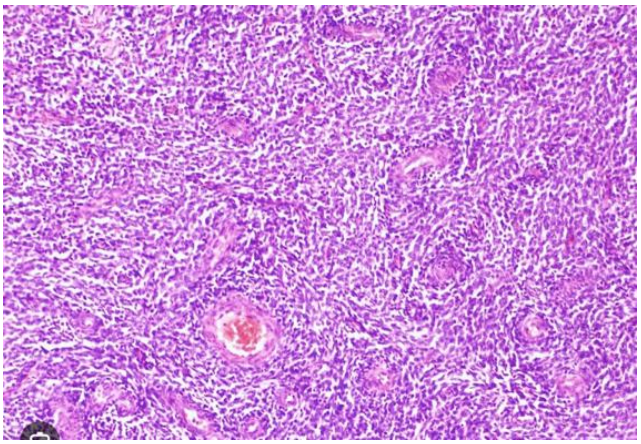

Figure 2: Histopathologic examination showing monotonous oval to spindle cells with minimal cytologic atypia, vesicular chromatin and scant cytoplasm

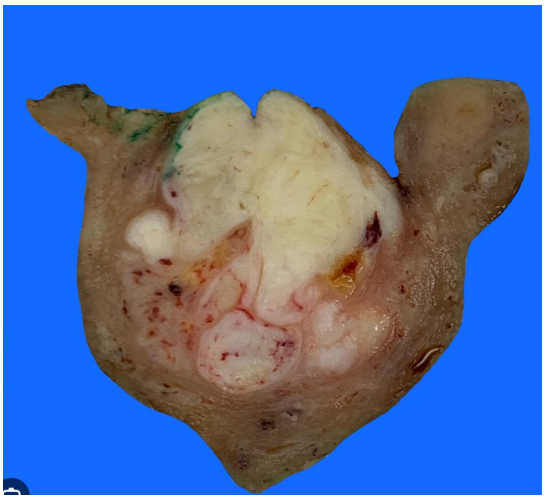

Figure 3: Gross examination of specimen showing poorly circumscribed soft yellow-tan to white nodules extending from the endometrium and invading into the myometrium

rare and this patient had only lower abdominal pain and lower abdominal mass.

Preoperative diagnosis is often misleading and around $3/4^{th}$ of the cases are misdiagnosed as benign leiomyoma. Endometrial curettage and HPE couldn't aid in diagnosis due to its likeliness towards normal endometrium.[8] Almost all the imaging procedures lack diagnostic accuracy and often misleading. Hence the absolute diagnostic method is histopathology combined with immunohistochemistry of the hysterectomy specimen.[9] Positive labelling for CD10, PR, ER with negativity for CD117, CD34 is a confirmative diagnosis for low grade ESS.

The management depends on the severity of the disease. Since it occurs more often in postmenopausal women, total abdominal hysterectomy with bilateral salpingo-oophorectomy is commonly suggested. Low-grade ESS had good prognosis of around 60% survival rate in next 5 years and the rate decreases to 25% if the condition is high grade. A study done by Behtash et al., showed that 96% of LGESS patients had survived in the next 5 years.[10]

4. Conclusion

ESS is an uncommon malignant tumor, presenting as unusual uterine bleeding in perimenopausal women. Although occasional, diagnosis of endometrial stromal tumors should be considered whenever a peri-menopausal women comes with lower abdominal pain and lower abdominal mass. By reporting our case, we wish to emphasise the prerequisite to have this tumour as a differential diagnosis and the importance of Contrast Enhanced MRI and LDH which could have diagnosed this tumour even in the absence of abnormal uterine bleeding. This case validates the significance of a thorough clinical examination and investigations. Total abdominal hysterectomy with bilateral salpingo oophorectomy with Adjuvant therapy is the definitive management for this condition.

5. Source of Funding

None.

6. Conflict of Interest

None.

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