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# ENDODERMAL STROMAL NODULE: A RARE CASE REPORT.

KEY WORDS:

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**ABSTRACT**

Endometrial stromal nodule (ESN) is the least common of the endometrial stromal tumors. They are rare neoplasms which are diagnosed in most instances by light microscopy. Although such nodules are benign, hysterectomy has been considered the treatment of choice to determine the margins of the tumor required for diagnosis and to differentiate it from invasive stromal sarcoma Whose prognosis is totally different. We report a case of a 50 years old woman, with presurgical diagnosis of adnexal mass or uterine tumor. She underwent an exploratory laparotomy with total abdominal hysterectomy. Pathologic examination revealed an endometrial stromal nodule. Through this observation, we insist on the fact that the ESNs are rare and benign entities which must be differentiated from the other invasive malignant stromal tumors; this can change the final prognosis.

## INTRODUCTION

Tumors arising from the endometrial stroma are exceedingly uncommon mesenchymal growths found within the uterus, exhibiting cytological and architectural characteristics akin to those of endometrial stromal cells.[2] Delineating the classification of endometrial stromal tumors poses a challenging and intricate task.[3,4] The recent WHO classification of tumors concerning the breast and female genital organs delineates uterine stromal neoplasms into three distinct categories: benign endometrial stromal nodules (ESNs), low-grade endometrial stromal sarcomas (LGESS), and undifferentiated endometrial sarcomas (UES).[5] ESNs, while cytologically resembling low-grade stromal sarcomas, are discernible by their well-defined, expansive margins and are typically considered clinically benign. Conversely, UES represents a rare yet highly malignant sarcoma devoid of evident endometrial stromal differentiation.[2] This study outlines a case involving a stromal nodule in a patient who underwent total abdominal hysterectomy, emphasizing the importance of carefully distinguishing endometrial stromal nodules from other stromal sarcomas, as this differentiation can significantly impact the final prognosis.

## BACKGROUND

Endometrial stromal nodules represent a rare subtype within the spectrum of endometrial stromal tumors (ESTs). ESTs constitute a mere 3% of all uterine neoplasms and are typically identified through light microscopy in the majority of cases.[6] One-fourth of these tumors manifest as endometrial stromal nodules. They can occur across a wide age range, spanning from 31 to 86 years, with a mean age of 54 years.[7]

In latest World health organization classification (2014), ESTs are divided into:[8]

- Endometrial stromal nodule (ESN) - least common
- Low grade endometrial stromal sarcoma (LG-ESS)
- High grade endometrial stromal sarcoma (HG-ESS)
- Undifferentiated endometrial sarcoma (USS).

Based on their histologic appearance, ESN and low-grade ESS fall in the lower end of the spectrum of ESTs. ESN is cytologically similar to low grade sarcomas. Both are composed of a diffuse growth of uniform small blue cells with scant cytoplasm and oval to spindle nuclei that resemble endometrial stromal cells of proliferative endometrium, that grow in sheets and focally whorl around arterioles.

ESN is differentiated from LG-ESS by it's an expansile but non infiltrative and smooth margins in contrast with an irregular nodular growth in endometrium with varying degrees of myometrial permeation as well as extension to extrauterine veins and lymphatics seen in the latter.

Microscopically, myometrial infiltration if present, can be seen as protrusions less than 3 in number of 3 mm or less with no angiolymphatic invasion and minimal mitotic activity (<3/10 HPF) 3 in a stromal nodule. Tumours with myometrial infiltration >3 mm and >3 in number would be categorized as LG-ESS.

High grade endometrial stromal sarcomas and undifferentiated endometrial stromal sarcomas show destructive infiltrative growth into the endometrium or myometrium or both. Microscopically, marked nuclear atypia is characteristic of high-grade ESS and UES with mitotic rate >10/10 HPF. The non-infiltrative border of stromal tumours is the single most important criterion for the diagnosis. Histological examination of the uterus is the most accurate method to make an appropriate diagnosis, thus hysterectomy being the treatment of choice in benign as well as malignant tumours. In this study we describe a young nulliparous patient who underwent conservative fertility preserving surgery and resection of mass which was postoperatively diagnosed as endometrial stromal nodule.

### Case Study

Mrs. ABC, 50 years old, P2L2 (all FTND), tubectomised, presented with dull pain and discomfort in the lower abdomen that had lasted for several weeks, gradual distension of abdomen and constipation for 2 months. The patient had her Menarche at the age of 14, and the menstrual cycles were regular without abnormal uterine bleeding until she was postmenopausal for 3 years.

**Per Abdominal Examination:** The abdominal examination revealed the abdominal pelvic painless mass reaching the

umbilical point, some parts of this mass were soft in consistency but the mass dependence on the uterus was not established; the rest of examination was without particularity. No tenderness, flank fullness present.

**Per Speculum Examination:** Anterior lip of the cervix was ballooned with cervical erosion. Posterior lip of cervix not seen. Vagina was healthy.

**PerVaginal Examination:** Posterior lip of cervix was high up. Firm mass of around 20 cm x 20 cm was felt. Bilateral fornices free non tender.

Laboratory investigations including serum CA 125, T3, T4, TSH were normal

USG Findings: 20.3 x 16.3 x 20.2 cm (volume – 3520 ml)Large soft tissue lesion with small cystic areas within. No evidence of internal vascularity.

Right side predominant, s/o? ovarian mass mostly of benign origin.

No free fluid in abdomen and pelvis.

The uterus and both ovaries not adequately visualized due to compression/displacement by the lesion.

**MRI Findings**
22.5 x 12.5 x 19.5 cm enhancing lesion in pelvis extending into the abdominal cavity the lesion causes displacement of uterus towards left side. Right ovary not identified separately from lesion. There is superior displacement of bowel loops. Anteriorly it is causing compression of urinary bladder. Uterus appears normal. The endometrium is linear and regular. The left ovary is normal in size and shape. Possibilities: Right ovarian neoplasm / Large sub serosal uterine fibroid.

**Plan of Action:**
Exploratory laparotomy with total abdominal hysterectomy with bilateral salpingo – oophorectomy

**Intra Op Findings**
Normal size uterus sitting on a huge mass (? Cervical fibroid) of size 20 x 20 cm. The gross inspection of hysterectomy specimen revealed a well-circumscribed yellow fleshy tumor measuring 20 x 20 cm that appears within the myometrial layer that resembled a leiomyoma. Bilateral fallopian tubes and ovaries grossly normal. bilateral round ligaments stretched over the mass.

No free fluid in the peritoneal cavity

Retroperitoneal structures grossly normal EBL- 1050 ml 2-pint PRBC given intraoperatively Mass weighed around 8.130 kg Peritoneal wash fluid was sent for cytology Mass sent for histopathology along with uterus, bilateral ovaries and fallopian tubes.

## DISCUSSION

Endometrial stromal tumours are relatively uncommon neoplasms of the uterine corpus, with an estimated annual incidence of around 2 per million women. [9,10,11,12] Diagnosis typically relies on light microscopy. Although the existence of circumscribed benign neoplastic proliferations of endometrial stromal cells, termed "endometrial stromal nodules," has been recognized for some time, literature on these lesions remains limited. These tumours, including ESNs, can resemble highly cellular leiomyomas and precede the availability of antibodies that aid in distinguishing EST from smooth muscle tumours, as well as in delineating the endometrial stromal and smooth muscle components in EST with smooth muscle metaplasia. Dionigi recently published a series of 50 cases, including ESTs with entirely circumscribed margins or limited focal infiltration at their periphery, and identified only four endometrial stromal nodules. Similarly, Amanjit reported five cases of EST out of 1261 endometrial neoplasms, with one case diagnosed as an endometrial stromal nodule. [13,14,15]

Endometrial stromal nodules are defined as well-circumscribed endometrial stromal tumours, although focal irregularities or finger-like projections into the adjacent myometrium are acceptable if none exceed 2 to 3 mm. [16,17]

ESNs, like other uterine neoplasms of stromal origin, primarily occur in peri- and postmenopausal women. Clinical presentation is often nonspecific, with patients presenting symptoms such as vaginal bleeding leading to anaemia, pelvic or abdominal pain or discomfort, or being asymptomatic. In our case, the patient experienced lower abdominal discomfort, pain, and distension without menorrhagia. The most common preoperative diagnoses are leiomyoma and adnexal masses. [9,18] Due to the age of most patients, hysterectomy is usually necessary for thorough evaluation of tumour margins, crucial for distinguishing benign stromal nodules from stromal sarcomas. However, unlike stromal sarcomas, patients with stromal nodules typically remain disease-free post-hysterectomy, with no noted recurrences. [17,18]

In cases where fertility preservation is desired, diagnostic imaging and hysteroscopy may be used for tumour growth monitoring. Hormonal therapy with local excision may be successful in some cases. Notably, hormonal therapy has been successful in decreasing the size of low-grade endometrial stromal sarcomas, allowing for local excision and preservation of reproductive function. [18]

Macroscopically, endometrial stromal nodules typically present as solitary, well-delineated, round fleshy nodules with a yellow to tan sectioned surface. Most are intramural without apparent connections to the endometrium, although occasional tumours may be cystic, with necrosis and haemorrhage being rare.

Differential diagnosis of endometrial stromal nodules depends on microscopic findings, which typically show areas of epithelial-like structures resembling ovarian sex cord tumors. Stromal nodules have expansile, noninfiltrative margins that compress the surrounding endometrium and myometrium. Minor irregularities of the margin are common, but invasion of the surrounding myometrium indicates a stromal sarcoma rather than a stromal nodule. [19,20]

Endometrial stromal tumors with sex cord-like elements exhibit a polyphenotypic immunophenotype, often displaying a mixed epithelial-myoid phenotype with immunoreactivity for cytokeratin and actin, and in some cases, desmin. Immunostains for EMA are typically negative. Inhibin and CD99 immunoreactivity may be detected in epithelial-like structures in approximately one-third of these tumors. In our case, the tumor expressed CD10 and hormonal receptors, while immunostaining for AML (alpha smooth muscle), desmin, calretinin, cytokeratin AE1/AE3, and inhibin were negative. [21]

Endometrial stromal nodules with focal sex cord-like differentiation tend to relapse and metastasize. In the initial report by Clement and Scully, three out of five patients with follow-up experienced recurrences, and two died. [21]

## CONCLUSIONS

We describe a case of an endometrial stromal nodule in a patient. Currently, there is no dependable preoperative diagnostic procedure available to accurately identify this tumor. Clinical presentation tends to be nonspecific. The

primary treatment option is hysterectomy. Diagnosis is confirmed through microscopic examination. It's crucial to precisely determine the tumor margins to distinguish it from invasive stromal tumors. Endometrial stromal nodules are generally regarded as benign tumors, and the prognosis is typically excellent when the diagnosis is certain.

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