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Endometrial Stromal Nodule: A Rare Case Report

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Case Report

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Abstract

Endometrial stromal tumors represent less than 2% of uterine tumors, being among the least common neoplasms of the uterine body. They can be divided into four main categories: endometrial stromal nodule, low-grade endometrial stromal sarcoma, high-grade endometrial stromal sarcoma and undifferentiated uterine sarcoma. This is a case of a 50-year-old female patient, who reported the onset of the disease in March 2022, characterized by anemia secondary to abnormal uterine bleeding and an unusual increased in abdominal diameter. She decided to go to a foreign hospital, where paraclinics studies were indicated. On July 12, 2022, she presented sudden abdominal pain, diffuse, of moderate to severe intensity, stabbing type and abdominal tenderness. Reason why she came to our center. Anemia and leukocytosis are determined. Imaging studies report a voluminous mass, mixed density, well delimited. CT reports another lesion of 5 cm that corresponds to a unicameral cyst of the right ovary. Midline laparotomy and endometrial protocol was made, with satisfactory evolution of the patient. Histopathological exam reported ESN. Conclusion: Hysterectomy is the treatment of choice. The anatomopathological study is fundamental for its final diagnosis and differentiation of stromal sarcomas, since its prognosis, treatment and follow-up are different.

Keywords: Laparotomy; Endometrial Stromal Tumors; Benign Neoplasm

Introduction

Endometrial stromal tumors represent less than 2% of uterine tumors. They are classified into four main categories, which are currently recognized by the WHO:

- Endometrial stromal nodule (ESN).
- Low-grade endometrial stromal sarcoma (LGESS).
- High-grade endometrial stromal sarcoma and (HGESS).
- Undifferentiated uterine sarcoma (UUS) [1].

ESN is the least common of the endometrial stromal tumor [2,3].

ESN are similar to LGEE, their differentiation between them is through the biopsy. ESN can be differentiated from the rest, due to their benign nature, being well circumscribed, with absence or minimal invasion of the myometrium (≤ 3 mm) and without vascular invasion [2-4]. Due to the rarity of these tumors, there is limited information concerning it.

Case Presentation

A 50-year-old female patient, with a history of controlled hypothyroidism, who presented in March 2022, anemia secondary to abnormal uterine bleeding and an unusual increased in abdominal diameter. She decided to go to a

foreign hospital, where paraclinics studies were indicated. On July 12, 2022, she presented sudden abdominal pain, diffuse, from moderate to severe intensity, stabbing type. Concomitant episode of hypotension. Reason why she came to our center, where she was evaluated and admitted for emergency surgical resolution. Gynecologic history: 2 pregnancies, 1 delivery and 1 caesarean section; menarche 14 years; date of last period 6/6/2022, cycles of 10 days duration; She denies the use of oral contraceptives and intrauterine device.

Clinical Findings

Physical exam, patient was dehydrated. Vital signs: heart rate: 115 beats per min, respiratory rate: 22 breaths per min, blood pressure: 90/50 mmHg, SPO2: 96%. Abdominal distention due to a space-occupying lesion at the pelvic level, which exceeds the umbilical scar, approximately 25 x 15 cm, with regular and smooth edges, mobile, low bowel sounds, painful on deep palpation in all quadrants, with abdominal guarding and tenderness. Pelvic examination with normal external genitals, normotonic, hyperthermic, vagina, short neck, painful on bilateral lateralization, bulging posterior fornix and fetid vaginal discharge. Rest of physical exam without alterations.

Paraclinics Studies

Laboratory test, hemoglobin: 6.4 gr/dl; leukocytes: 16 103/mm3; neutrophil: 84%; c-reactive protein: 25mg/dl. Rest without alteration. The patient had studies prior to her arrival, CA-125: 9.13IU/ml. Computed tomography (CT) of Abdomen and pelvic with double contrast that reports a voluminous mass of 20 cm in diameter, mixed density, well delimited, that rejects the small intestine and colon. In addition, next to the mass described above, CT report another lesion of 5 cm that corresponds to a unicameral cyst of the right ovary (Figure 1).

Figure 1A: Transvaginal color doppler ultrasound.

Figure 1B: CT abdominal and pelvic with double contrast, axial view.

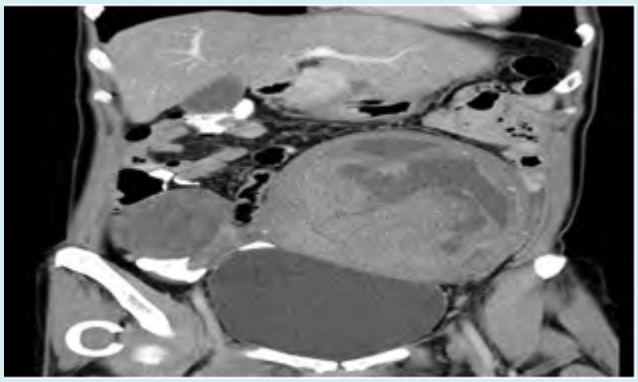

Figure 1C: CT abdominal and pelvic with double contrast, coronal view.

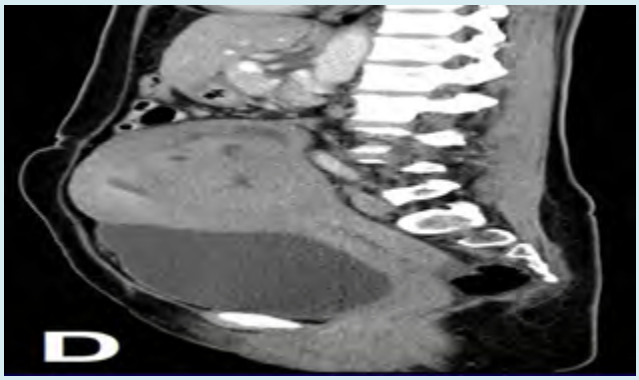

Figure 1D: CT abdominal and pelvic with double contrast, sagital view.

Cervical cytology and endometrial biopsy were negative for malignancy. She is admitted with a diagnosis of peritonitis of gynecological origin.

Treatment Performed

Midline laparotomy was made. Once in the abdominal cavity, a sample of the peritoneal fluid was taken, and it is reviewed. Subsequently, a total abdominal hysterectomy with bilateral salpingo-oophorectomy. Samples were taken from the peritoneal surface of the paracolic grooves and subphrenic spaces, infrasonic omentectomy, pelvic and para-aortic lymphadenectomy because intraoperative frozen biopsy couldn't be made. The findings were the following:

- 500 cc of peritoneal fluid, fetid.
- 20 x 10 cm, thick-walled, smooth-surfaced uterus.
- Right ovary 7 x 5 cm, cystic.
- Left ovary 4 x 3 cm, cystic.
- Pelvic and para-aortic nodes without macroscopic evidence of involvement.
- Rest without alterations.

Because of the intraoperative findings, the postoperative diagnosis of infected endometrial tumor and bilateral ovarian cysts is decided.

Follow-Up and Results

The patient received red blood cell transfusion during the surgery. Post-transfusion hematology test was performed, and normal results were obtained. Patient is discharged on the second postoperative day and evolves satisfactorily during the following two months of postoperative controls.

Histopathological examination reported a 2.5 kg uterus, with an ovoid lesion inside, soft, adhered to the posterior wall, 17 x 13 x 8 cm, without invasion of veins and lymphatic vessels, <3 mm infiltration of the myometrium, non-infiltrative tumor margins. The resection specimen confirmed the diagnosis of endometrial stromal nodule. Right ovary with serous cyst and left ovary with simple serous cyst. Sub phrenic peritoneum and paracolic grooves without significant histological lesion. Peritoneal fluid with the presence of ovoid cells, with little cytoplasm, monomorphic, without mitosis and without pleomorphism (Figure 2).

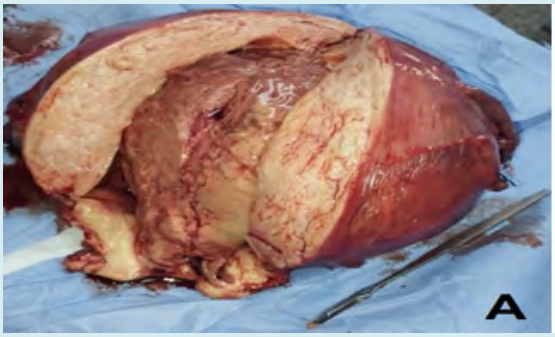

Figure 2A: Surgical specimen removed.

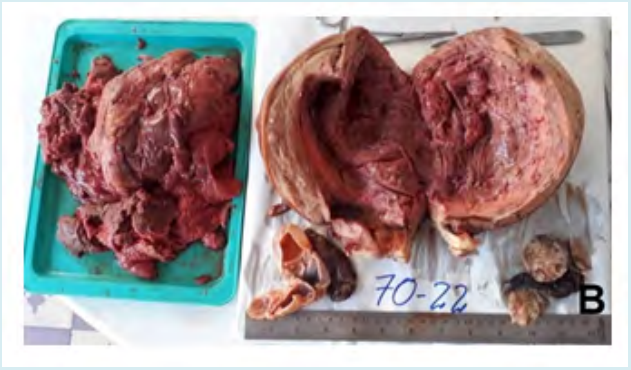

Figure 2B: Macroscopic finding in the histopathological exam of the lesion.

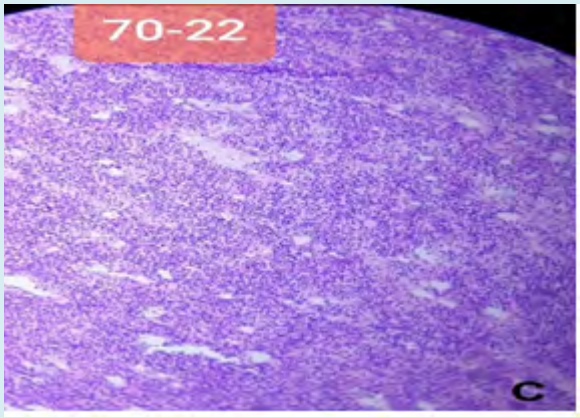

Figure 2C: Microphotograph HE-10x. Endometrial stromal nodule with ovoid cells, without mitosis and small myxoid lacunae.

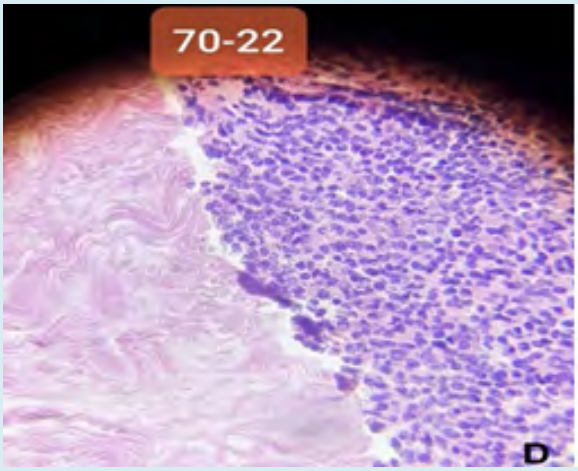

Figure 2D: Microphotography HE-10x. Left: myometrium, right: endometrial stromal nodule lesion that does not invade or infiltrate the myometrium. It is well defined.

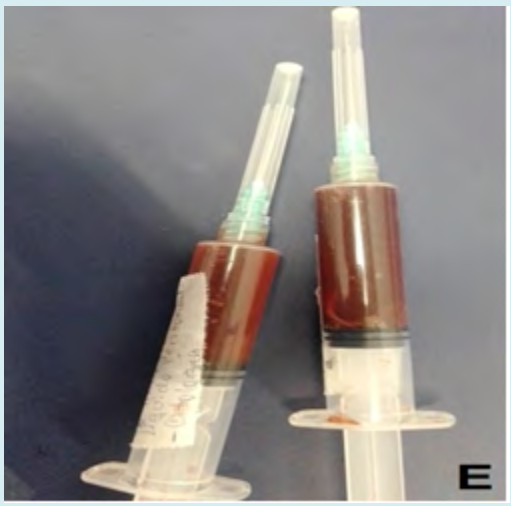

Figure 2E: Extracted peritoneal fluid.

Discussion

Endometrial stromal tumors are among the least common neoplasms of the uterine body, with an annual incidence of 2 per million women [2,3]. ESN occur mainly in peri- and postmenopausal women. The clinical manifestations are not specific, and it may be asymptomatic or present anemia secondary to abnormal uterine bleeding, abdominal or pelvic pain [2]. Although this pathology is benign, hysterectomy remains like 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 [2]. For a woman of reproductive age who wishes to preserve her fertility, imaging studies and hysteroscopy may be used to monitor tumor growth. In some cases, hormone therapy with local excision can be successful [2]. Until now, there are no immunohistochemical biomarkers that differentiate ESN from possible malignant sarcomas prior to hysterectomy [5]. Macroscopically, the tumor has characteristics of a well-demarcated, solitary, round nodule with a yellow to brown cut surface. The average diameter of the tumor is 4 cm (range 0.8 to 15 cm). Ours was 20 cm. About two-thirds are purely intramural, with no apparent connection to the endometrium. Occasionally, the tumors are cystic, while foci of necrosis and hemorrhage are rare [2].

Microscopically, ESNs are a proliferation of bland, uniform cells with oval nuclei and scanty cytoplasm, resembling endometrial stromal cells in the proliferative phase of the menstrual cycle. ESNs exhibit prominent arterioles and well-circumscribed expansive (non-infiltrative) margins. It may occasionally present with infiltrative margins to the myometrium ≤ 3 mm [6]. In a curettage specimen, the difference between ESN and LGESS is almost impossible, unless the tumor is very small and the margins can be fully evaluated [2]. ESNs can be differentiated from LG-ESS exclusively by the presence of pushing margins and lack of lymph-vascular invasion [6]. Immunohistochemistry, both ESN and LGESS demonstrate positive immunoreactive with CD10, but around 10-25% of ESN may be negative [3,4]. Other markers expressed by both are: estrogen and progesterone receptors, smooth muscle actin and vimentin [6]. Molecular classification is currently being used, where the most common genetic fusion expressed is JAZF1-SUZ12, found in half of the cases of ESN and LGESS. Recently, MEAF6-PHF1 has also been demonstrated in ESNs [6]. Patients with ESN after hysterectomy remain free of disease and without recurrences [2].

Conclusion

ESNs are rare and benign entities which must be differentiated from the other endometrial stromal sarcoma; this can change the final prognosis. Hysterectomy is the treatment of choice.

Conflict of Interests

The authors declare that they have no conflict of interest.

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