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Case Report

Low-grade endometrial stromal sarcoma, a rare uterine tumor: Case report ^{☆,☆☆}

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ABSTRACT

This case report describes a 45-year-old woman presenting with abnormal uterine bleeding and a cervical mass. Imaging and biopsy revealed low-grade endometrial stromal sarcoma (LGESS), emphasizing the importance of comprehensive evaluation for uterine masses. The report underscores the role of MRI and pathology in diagnosis, with immunohistochemical analysis helping confirmation. A multidisciplinary approach and vigilant follow-up are crucial for optimal management. The rarity of LGESS and its challenging diagnosis highlight the need for continued research to improve diagnostic and therapeutic strategies. Surgical intervention remains primary, but the optimal management approach is debated. This report indicates the necessity of a comprehensive approach to uterine mass evaluation and ongoing research for enhanced patient care.

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Introduction

Uterine masses are a common concern for women of reproductive age and can have various etiologies, including malignancy. Accurate diagnosis is crucial for prompt and appropriate management. This case report presents a 45-year-old female who presented with abnormal uterine bleeding and was found to have a cervical growth on pelvic examination. Fur-

ther imaging and biopsy confirmed the diagnosis of low-grade endometrial stromal sarcoma, emphasizing the importance of comprehensive imaging and biopsy in the evaluation of uterine masses and the role of immunohistochemical analysis in challenging cases. This case highlights the need for a multidisciplinary approach in the management of uterine masses and underscores the importance of vigilant follow-up to ensure optimal outcome.

Abbreviations: MRI, Magnetic Resonance Imaging; LGESS, Low-Grade Endometrial Stromal Sarcoma; H&E, Hematein & Eosin; IHC, Immunohistochemistry; DWI, Diffusion Weighted Imaging.

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Fig. 1 – Sagittal T2 view of the tumor on MRI.



Fig. 2 – Coronal T2 view of the tumor on MRI.

Case report

A 45-year-old female with no significant medical history presented to the Emergency Department with abnormal uterine bleeding leading to anemia. Her general examination was unremarkable, but a pelvic examination revealed an indurated vagina and a mass hanging from the cervix that was bulging into the external cervical orifice.

Further imaging with ultrasound and magnetic resonance imaging (MRI) revealed the cervical growth, measuring $44 \times 41 \times 54$ mm, isointense on T2 (Figs. 1 and 2), hyperintense on DWI (Fig. 3) and heterogeneously enhanced on postgadolinium T1 sequences (Fig. 4). However, imaging alone was not sufficient for a definitive diagnosis, and an endoscopy-guided biopsy was performed.

The biopsy revealed the need for complementary analysis with immunochemistry. The histopathological examination was initially inconclusive, but complementary immunohistochemistry showed positivity for CD10 and Estrogen Receptor and negativity for H-caldesmon. The Ki67 index of proliferation was low with scattered nuclear staining, leading to a final diagnosis of low-grade endometrial stromal sarcoma (Fig. 5).

To manage the case, a polypectomy with endometrial curettage was performed. The patient's treatment and follow-up will be closely monitored to ensure optimal outcome.

Discussion

Low-grade endometrial stromal sarcoma (LGESS) is a rare form of uterine sarcoma, accounting for only 2%-4% of all uterine sarcomas and 0.2% of all uterine malignancies [1]. It is characterized by a slow-growing and indolent nature, with an average time to recurrence of approximately 6 years and a 5-year

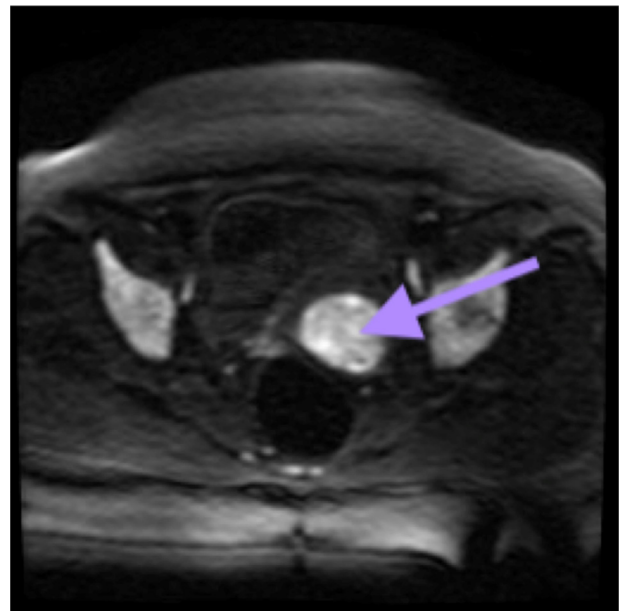


Fig. 3 – Axial DWI view of the tumor on MRI.

overall survival rate of approximately 80% [2]. The tumor often presents with atypical symptoms, such as abnormal uterine bleeding, making it a challenging diagnosis [3]. This highlights the importance of considering LGESS in the differential diagnosis of uterine masses and emphasizes the need for comprehensive imaging and biopsy techniques to confirm the diagnosis [2].

The diagnosis of LGESS is often confirmed through a combination of clinical, imaging, and pathological findings. Magnetic resonance imaging (MRI) is a valuable tool in the evalu-

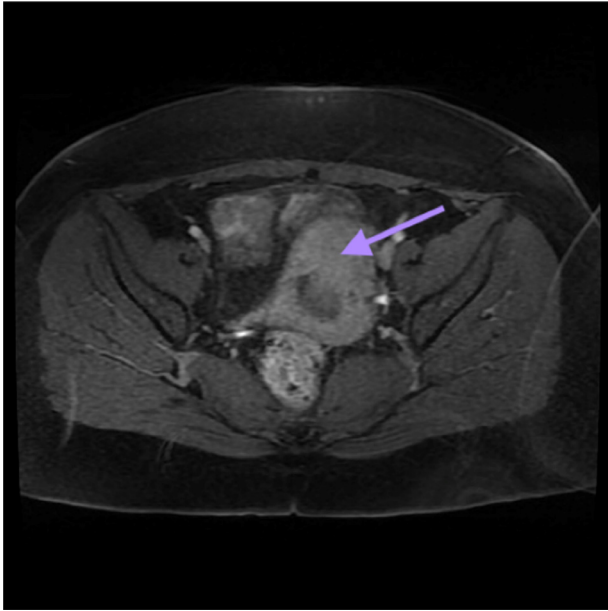


Fig. 4 – Axial T1 FS view of the tumor on MRI postgadolinium.

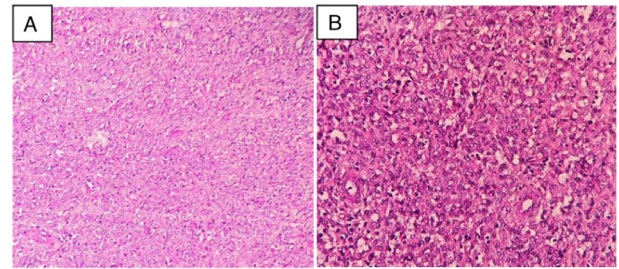
ation of uterine masses and can help to distinguish between benign and malignant tumors [4].

On MRI, LGESS can have many aspects but typically has a low T1 signal and ill-defined borders that are best shown on T2 sequences with a faint high-signal that suggests central necrosis [5]. It also has a strong and early enhancement post gadolinium with hyperintense signal on diffusion imaging (DWI) [5]. In our patient, the low T1 signal and the hyperintense DWI signal is present but the borders are pretty well defined on all sequences, which makes our case atypical. This polymorph aspect of LGESS explains the importance of pathology for diagnosis.

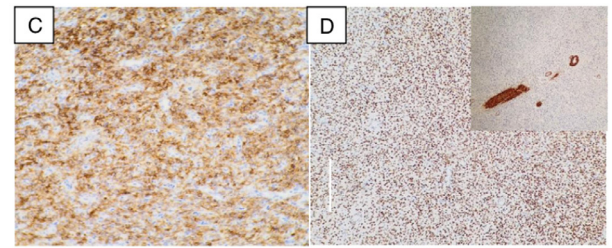
Endoscopy-guided biopsy and its histopathological examination are therefore critical in the confirmation of the diagnosis, with immunohistochemical analysis being a valuable adjunctive tool in challenging cases [3]. The positive expression of CD10 and estrogen receptor in this case, as well as the negative expression of H-caldesmon, supports the diagnosis of LGESS [3,6].

It is important to report cases of LGESS as they provide valuable information and insight into the diagnosis, management, and outcome of this rare form of uterine sarcoma that can metastasize [7]. By sharing experiences and knowledge, healthcare professionals can gain a better understanding of the challenges involved in diagnosing and treating LGESS and improve the care provided to patients. Moreover, reporting cases of LGESS can also contribute to the advancement of research in this field and lead to the development of better diagnostic and therapeutic approaches.

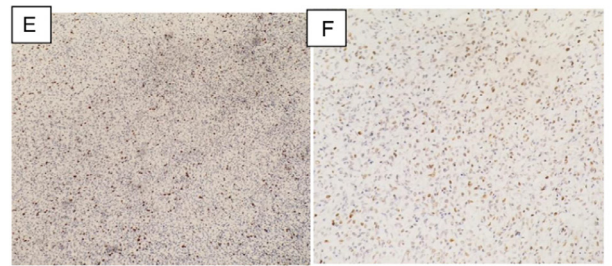
The management of LGESS is primarily surgical, with the goal of complete removal of the tumor [8]. In some cases, adjuvant radiation therapy may also be recommended to reduce the risk of recurrence [8]. However, the optimal management



Monotonous spindle to oval cells with minimal nuclear atypia (H&E)
A: 100x
B: 200x



Immunohistochemistry: the cells are typically positive for CD10(C) and estrogen receptor (D)
Note the negativity of H-caldesmon with positive internal control (encart)



Immunohistochemistry: Ki67 index of proliferation is low (E)
Wildtype P53, scattered nuclear staining (F)
(IHC 100x)

Fig. 5 – Microscopic studies of the tumor.

strategy for LGESS is still a topic of debate, and further research is needed to determine the most effective approach.

In conclusion, reporting cases of low-grade endometrial stromal sarcoma is of great importance as it helps to increase awareness of this condition, improve diagnosis, and management, and contribute to the advancement of research in this field. The diagnosis of LGESS is challenging, but with a multidisciplinary approach, including clinical examination, imaging, biopsy, and immunohistochemical analysis, it can be accurately confirmed. Vigilant follow-up is essential to monitor for recurrence and ensure optimal outcome.

This case report serves as a reminder of the need for a comprehensive approach in the evaluation and management of uterine masses and the importance of continued research in the field of LGESS.

Patient consent

I hereby affirm that written and informed consent for the publication of the case has been obtained from the patient(s) involved. This statement is intended to appear in the article. I want to emphasize that the agreement with the patient(s) or their representative(s) will not be submitted but shall be retained for my own records.

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