Int J Clin Exp Pathol 2026;19(5):209-214
www.ijcep.com /ISSN:1936-2625/IJCEP0171234

Case Report

Myolipoma of the uterus with bizarre-nucleated cells: a case report and literature review

Li Zhang, Chuanling Niu

Department of Pathology, East Sichuan Hospital of Sichuan Provincial People's Hospital and Dazhou First People's Hospital, Dazhou 635000, Sichuan, China

Received January 4, 2026; Accepted April 9, 2026; Epub May 15, 2026; Published May 30, 2026

Abstract: Background: Myolipoma is a rare benign mesenchymal tumor composed of mature adipose tissue, smooth muscle bundles and blood vessels, which predominantly occurs in the retroperitoneum, inguinal region and abdominal wall. Primary uterine involvement is extremely uncommon, and the presence of bizarre-nucleated cells within such tumors is even rarer. The morphological features of these cells may mimic malignant neoplasms, posing great challenges for clinical and pathological diagnosis. Methods: We report a 61-year-old female patient with imaging and ultrasonographic findings suggestive of a uterine space-occupying lesion. Histopathological examination and immunohistochemical staining were performed on the surgically resected specimen, with the detected markers including smooth muscle actin (SMA), Desmin, HMB45, Melan A, S-100 and Ki-67. Results: Pelvic magnetic resonance imaging (MRI) revealed a well-demarcated heterogeneous signal mass in the anterior wall of the uterine corpus, consistent with the manifestations of a benign tumor containing adipose components. Gross examination showed a well-circumscribed intramural mass of the uterus with a grayish-yellow and heterogeneous cut surface. Microscopically, mature adipocytes and benign smooth muscle cells were scattered with bizarre-nucleated cells characterized by markedly enlarged and irregular nuclei; no mitotic figures or necrotic foci were identified. Immunohistochemically, the smooth muscle component showed diffuse and strong positivity for SMA and Desmin, while HMB45, Melan A and S-100 were all negative. The Ki-67 proliferation index was extremely low (<1%), confirming the diagnosis of uterine myolipoma with bizarre-nucleated cells. Conclusion: Uterine myolipoma with bizarre-nucleated cells is an exceedingly rare benign tumor, and no recurrence or metastasis was observed during postoperative follow-up, indicating its indolent biological behavior. Accurate diagnosis requires the combination of imaging features, adequate sampling, meticulous histological evaluation and targeted immunohistochemical detection to distinguish it from leiomyosarcoma, liposarcoma and perivascular epithelioid cell tumor (PEComa).

Keywords: Uterine neoplasms, myolipoma, bizarre-nucleated cells, case report, literature review

Introduction

Myolipoma, a rare benign mesenchymal tumor first described by Weiss and Rao in 1992, is mainly composed of mature adipose tissue, smooth muscle bundles and thick-walled blood vessels. It predominantly arises in the retroperitoneum, inguinal region and abdominal wall, and primary uterine myolipoma is extremely uncommon with only a small number of case reports published domestically and internationally [1, 2]. In recent years, myolipomas with bizarre-nucleated cells have attracted increasing attention from pathologists because their morphological features are easily misdiagnosed as malignant lesions, which further increases the difficulty of clinical diagnosis [3, 4].

Current research on myolipoma remains limited, and the reported cases are mostly concentrated in deep soft tissues. As an organ with a high incidence of smooth muscle tumors, the uterus harbors a variety of mesenchymal neoplasms, among which leiomyoma is the most common benign tumor and leiomyosarcoma is the most frequent malignant type [5, 6]. Due to the unique histological composition and extremely low incidence of myolipoma, cases with bizarre-nucleated cells are often clinically misdiagnosed as leiomyosarcoma or other malignant tumors, thus in-depth investigation and

https://doi.org/10.62347/XATC8124

analysis of such lesions are urgently needed [7, 8].

Two main hypotheses exist regarding the histogenesis of myolipoma: one holds that it is a hamartomatous lesion, and the other proposes that it is a benign tumor with unique differentiation potential [9, 10]. Negative immunoreactivity for HMB45 and Melan A is helpful in distinguishing myolipoma from PEComa, providing a key basis for diagnosis [11, 12]. Aiming at the gaps in existing research, this study conducts a detailed pathological analysis of a case of uterine myolipoma with bizarre-nucleated cells combined with imaging data and relevant literature review, to improve the understanding of this rare tumor among clinicians and pathologists.

In this study, reagents from Hangzhou ZSGB-BIO Technology Co., Ltd. were used to perform detailed pathological examination and immunohistochemical analysis. We focused on the three histological components (adipose tissue, smooth muscle and blood vessels) and characteristic bizarre-nucleated cells of the tumor, and made a comprehensive judgment combined with imaging findings to ensure accurate diagnosis and differential diagnosis [4, 13]. It is expected that this case report can provide a reference for the diagnosis of similar cases and highlight the importance of identifying myolipoma and its subtypes to avoid clinical overtreatment.

Primary uterine myolipoma with bizarre-nucleated cells is an extremely rare benign tumor, and its diagnosis relies on imaging evaluation, meticulous morphological observation and appropriate immunohistochemical detection. In-depth exploration of this lesion is expected to provide new ideas for research in this field and promote the upgrading of clinical understanding of the disease.

Case report

Clinical data and imaging findings

A 61-year-old female was admitted to the hospital with a 15-year history of vulvar pruritus and a 1-year history of a vulvar mass. The patient first found the vulvar mass 1 year ago but did not seek medical treatment in a timely manner, and was later referred to the outpatient department of our hospital for further diagnosis and treatment. She had no special past medical history or family genetic history. Gynecological examination revealed that her uterus was the size of 2-weeks of gestation with an irregular shape and no tenderness; no obvious abnormalities were palpable in the bilateral adnexal regions, and an irregular soft tissue mass was seen in the vulva. Ultrasonography suggested a uterine space-occupying lesion and a vulvar soft tissue mass, and further pelvic MRI was completed to clarify the nature of the lesion (**Figure 1A**, **1B**). The patient subsequently underwent total hysterectomy and vulvectomy.

Gross pathological examination

The total hysterectomy specimen measured approximately 18 cm × 17 cm × 4 cm. Longitudinal incision revealed a grayish-white firm nodule with a maximum diameter of about 7 cm in the uterine myometrium, with clear boundaries and no capsule. Comprehensive sampling of the remaining uterine wall identified a second tumor with a maximum diameter of about 1.4 cm. The cut surface was grayish-yellow, with partial areas showing pale yellow adipose-like appearance and partial areas showing pale white smooth muscle-like appearance, with a moderately soft to firm texture. No obvious necrosis or hemorrhagic foci were observed (**Figure 2A**).

Microscopic examination (hematoxylin-eosin staining)

At low magnification, the tumor was well-demarcated from the surrounding uterine myometrium and exhibited an expansile growth pattern (**Figure 2B**, scale bar = 200 μm). Mature adipose tissue accounted for approximately 60%-70% of the tumor volume, with well-differentiated and uniformly sized adipocytes. Smooth muscle bundles accounted for 30%-40%, composed of well-differentiated spindle cells arranged in interlacing bundles with eosinophilic cytoplasm (**Figure 2C**, scale bar = 50 μm). Bizarre-nucleated cells were scattered within the smooth muscle bundles, characterized by markedly enlarged, hyperchromatic and highly irregular nuclei with a lobulated or multinucleated giant cell-like morphology and coarse chromatin (**Figure 2D**, scale bar = 50 μm). No pathological mitotic figures were found

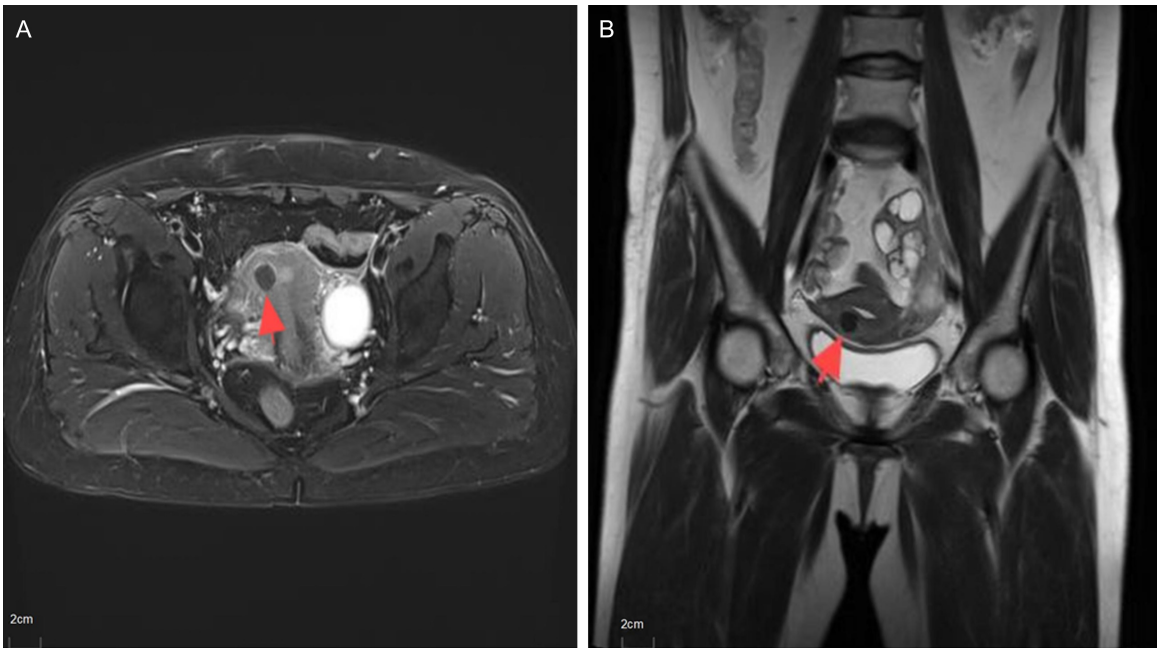

Figure 1. Imaging findings of uterine myolipoma with bizarre-nucleated cells. A: Axial view of plain pelvic MRI showing a well-demarcated heterogeneous signal mass in the anterior wall of the uterine corpus with adipose and soft tissue signal components (marked by red arrows); B: Sagittal view of pelvic MRI fat-suppressed sequence showing suppressed fat signals in the uterine mass, confirming that the lesion contains adipose and smooth muscle tissue components, consistent with the typical imaging characteristics of uterine myolipoma (marked by red arrows).

(mitotic count <1 per 10 high-power fields), and no tumor coagulative necrosis or map-like necrosis was observed. There was no obvious cellular crowding or dysplasia around the bizarre-nucleated cells.

Immunohistochemical staining

Four μm formalin-fixed paraffin-embedded tissue sections were prepared, and immunohistochemical staining was performed using the streptavidin-peroxidase (SP) method, with all reagents purchased from Hangzhou ZSGB-BIO Technology Co., Ltd. The secondary antibody was goat anti-mouse/rabbit IgG polyclonal antibody at a dilution ratio of 1:200. The dilution ratios of each primary antibody were as follows: SMA 1:200, Desmin 1:150, HMB45 1:100, Melan A 1:100, S-100 1:300, Ki-67 1:200, CD34 1:200, and STAT6 1:150. The staining results were as follows: adipocytes: S-100 (+) (**Figure 2E**, scale bar = 50 μm); smooth muscle cells (including bizarre-nucleated cells): diffuse and strong positivity for Desmin (**Figure 2F**, scale bar = 50 μm) and SMA (**Figure 2G**, scale bar = 50 μm); bizarre-nucleated cells: negative for HMB45 (**Figure 2H**, scale bar = 50 μm) and Melan A (**Figure 2I**, scale bar = 50 μm); tumor cells: CD34 (positive in vascular endothelium, negative in tumor cells), STAT6 (-); the Ki-67 proliferation index was <1% (bizarre-nucleated cells were also negative) (**Figure 2J**, scale bar = 50 μm).

Pathological diagnosis

Myolipoma with bizarre-nucleated cells (in the uterine myometrium).

Postoperative follow-up

The patient had an uneventful postoperative recovery. Gynecological examination and pelvic ultrasonography were performed at 6 months and 1 year after surgery, and no signs of tumor recurrence were found, confirming the indolent biological behavior of the tumor.

Discussion

Myolipoma is a rare benign mesenchymal tumor composed of mature adipose tissue, benign smooth muscle cells and blood vessels, which mainly occurs in the retroperitoneum, inguinal region and abdominal wall. Primary

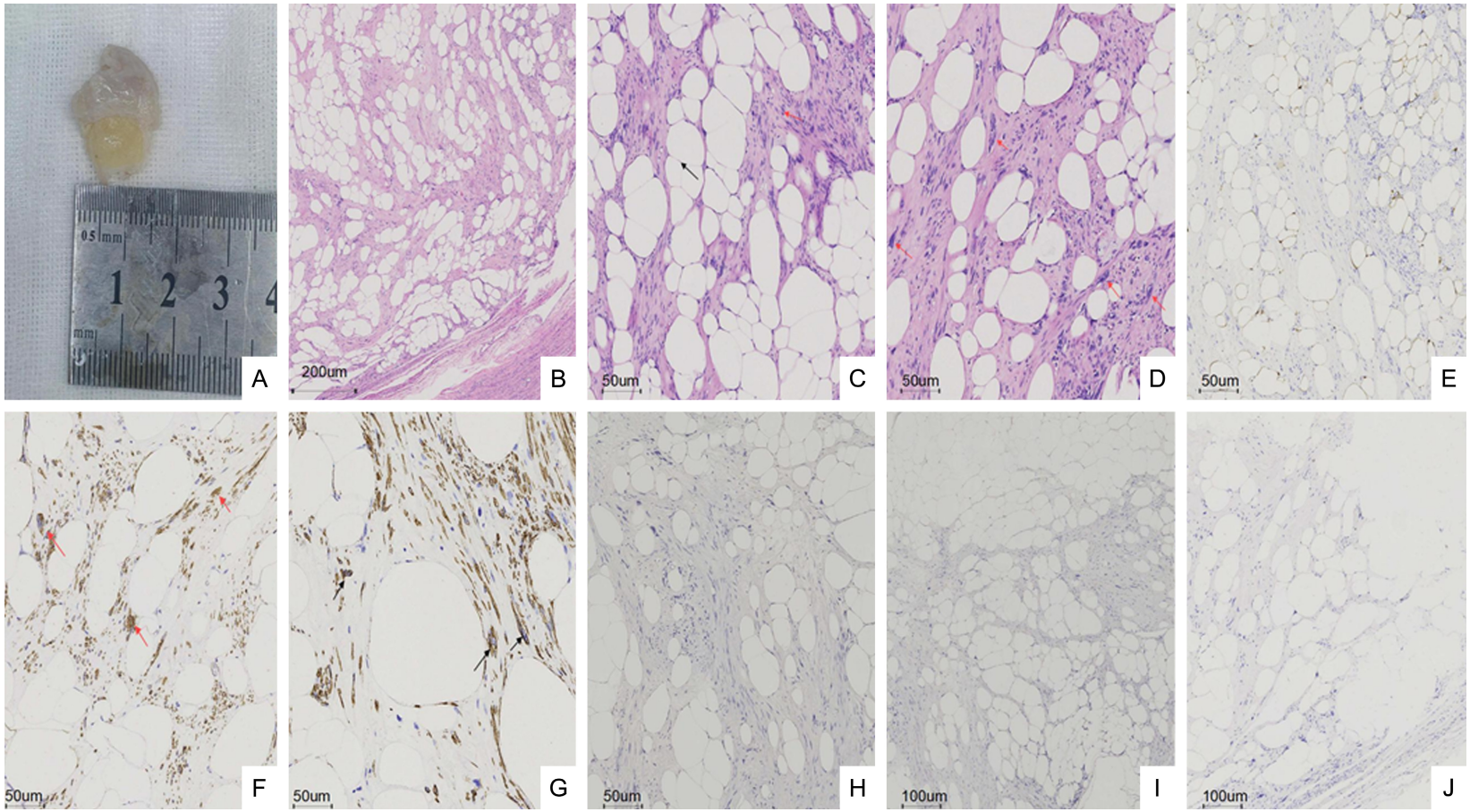

Figure 2. Pathological morphological and immunohistochemical findings of uterine myolipoma with bizarre-nucleated cells. A: Gross appearance of the uterine tumor with a grayish-yellow, heterogeneous cut surface showing distinct adipose-like and smooth muscle-like areas, clear boundaries, and no evidence of necrosis or hemorrhage; B: Low-magnification HE staining (scale bar = 200 μm) showing the tumor is well-demarcated from the surrounding uterine myometrium with an expansile growth pattern; C: Medium-magnification HE staining (scale bar = 50 μm) showing the tumor is mainly composed of mature adipose tissue (marked by black arrows) and interlacing smooth muscle bundles (marked by red arrows); D: High-magnification HE staining (scale bar = 50 μm) showing scattered bizarre-nucleated cells with enlarged, hyperchromatic and irregular nuclei in the smooth muscle bundles (marked by red arrows); E: S-100 immunohistochemical staining (scale bar = 50 μm) showing strong cytoplasmic positivity in mature adipocytes; F: Desmin immunohistochemical staining (scale bar = 50 μm) showing diffuse and strong cytoplasmic positivity in smooth muscle cells and bizarre-nucleated cells (marked by red arrows); G: SMA immunohistochemical staining (scale bar = 50 μm) showing diffuse and strong cytoplasmic positivity in smooth muscle cells and bizarre-nucleated cells; H: HMB45 immunohistochemical staining (scale bar = 50 μm) showing negative expression in all tumor cells including bizarre-nucleated cells; I: Melan A immunohistochemical staining (scale bar = 100 μm) showing negative expression in all tumor cells including bizarre-nucleated cells; J: Ki-67 immunohistochemical staining (scale bar = 100 μm) showing an extremely low proliferation index of the tumor with no positive expression in bizarre-nucleated cells.

uterine involvement is extremely uncommon with few literature reports [1, 2]. The presence of bizarre-nucleated cells in the smooth muscle component of the tumor is even rarer, and these cells are large with irregular morphology, which are easily misjudged as malignant features, further increasing the diagnostic difficulty [4, 8].

This study reports a case of uterine myolipoma with bizarre-nucleated cells and conducts an analysis combining pelvic MRI imaging features, pathological morphology and immunohistochemical results. Standardized reagents from Hangzhou ZSGB-BIO Technology Co., Ltd. were used in the study to clarify the pathological features and immunohistochemical phenotype of the tumor. Combined with imaging findings, it provides a key basis for accurate diagnosis and reduces the risk of misdiagnosis as leiomyosarcoma or other malignant tumors [3, 9]. By sorting out the existing literature, this study aims to improve the cognition of clinicians and pathologists on this rare lesion, and emphasize the importance of integrating imaging findings, meticulous evaluation of morphological features and immunohistochemical results in the diagnostic process.

The characterization of this tumor reveals its unique biological properties. Although myolipoma is a benign lesion, the presence of bizarre-nucleated cells is prone to malignant misdiagnosis. These cells are mostly degenerative changes or morphological variations of cells rather than malignant indicators, which confirms the benign nature of the tumor and also highlights the necessity of meticulous histological evaluation and immunohistochemical detection [2, 13]. Meanwhile, the Ki-67 proliferation index <1% indicates extremely low proliferative activity of the tumor, consistent with the biological behavior of benign tumors. Preoperative MRI also suggested that the lesion was a benign space-occupying lesion containing fat, which was mutually confirmed with the pathological results, providing an important basis for interpreting tumor characteristics and guiding subsequent diagnostic strategies.

From the perspective of cellular biology and tumor microenvironment, the presence of bizarre-nucleated cells may affect the intercellular interaction of tumor cells and the surrounding microenvironment, thereby regulating tumor progression. Despite concerns about malignant potential, no recurrence or metastasis was found during 1-year follow-up, indicating that bizarre-nucleated cells in this type of tumor do not represent malignant biological behavior [4, 6]. This finding refines the understanding of relevant biological processes and also highlights the importance of integrating imaging, cellular behavior and microenvironmental factors in tumor diagnosis.

In terms of immunohistochemistry, the phenotype of the smooth muscle component is closely correlated with bizarre-nucleated cells. Bizarre-nucleated cells do not express melanocyte markers (HMB45, Melan A), while smooth muscle cells show diffuse and strong expression of smooth muscle-specific markers (SMA, Desmin) [1, 3]. This distinct immunophenotype is a key basis for differential diagnosis, helping pathologists distinguish myolipoma from potentially malignant tumors such as leiomyosarcoma and PEComa. Therefore, clarifying the immunological characteristics of the tumor and making a comprehensive judgment combined with imaging findings are of great clinical significance for formulating individualized diagnosis and treatment plans and monitoring disease progression.

This study has certain limitations: it is a single-case study with a small sample size; in addition, the lesion is extremely rare with few relevant literature cases, which brings challenges to the generalization of the research conclusions in clinical practice. Furthermore, the lack of long-term follow-up data makes it impossible to clarify the biological behavior and prognostic factors of the tumor in different populations. In the future, multi-center and large-sample cohort studies need to be carried out to accumulate more comprehensive clinical, imaging and pathological data, so as to further improve the cognition and diagnosis and treatment level of this rare lesion.

Conclusion

Uterine myolipoma with bizarre-nucleated cells is an extremely rare benign uterine neoplasm. Accurate identification of its imaging, clinical and pathological features is the key to avoiding misdiagnosis and eliminating unnecessary overtreatment. Preoperative MRI evaluation of

lesion nature, adequate intraoperative sampling, meticulous postoperative morphological evaluation, combined with targeted immunohistochemical detection using standardized reagents, can effectively distinguish this tumor from malignant neoplasms such as leiomyosarcoma, liposarcoma and PEComa. This study enriches the clinical, imaging and pathological data of such rare uterine tumors, and emphasizes the necessity of strengthening the training of clinicians and pathologists and improving the sensitivity and specificity of rare lesion identification, ultimately reducing unnecessary surgical interventions and alleviating patients' psychological burden. More clinical cases need to be accumulated for long-term follow-up in the future to further clarify the natural course and prognostic factors of this rare tumor.

Disclosure of conflict of interest

None.

Address correspondence to: Chuanling Niu, Department of Pathology, East Sichuan Hospital of Sichuan Provincial People's Hospital and Dazhou First People's Hospital, Dazhou 635000, Sichuan, China. Tel: +86-17753799711; E-mail: ChuanlingNiu@163.com

References

[1] Ünver NU, Acikalin MF, Öner Ü, Ciftci E, Ozalp SS and Colak E. Differential expression of P16 and P21 in benign and malignant uterine smooth muscle tumors. Arch Gynecol Obstet 2011; 284: 483-490.

[2] Hubelé F, Averous G, Rust E, Imperiale A and Namer IJ. FDG-PET/CT findings of atypical (bizarre/symplastic) uterine leiomyoma in a patient with abdominal leiomyosarcoma. Radiol Case Rep 2015; 7: 670.

[3] Pang SJ, Li CC, Shen Y, Liu YZ, Shi YQ and Liu YX. Value of counting positive PHH3 cells in the diagnosis of uterine smooth muscle tumors. Int J Clin Exp Pathol 2015; 8: 4418-4426.

[4] Zhang Q, Kanis MJ, Ubago J, Liu D, Scholtens DM, Strohl AE, Lurain JR, Shahabi S, Kong B and Wei JJ. The selected biomarker analysis in 5 types of uterine smooth muscle tumors. Hum Pathol 2018; 76: 17-27.

[5] Ip PP, Tse KY and Tam KF. Uterine smooth muscle tumors other than the ordinary leiomyomas and leiomyosarcomas: a review of selected variants with emphasis on recent advances and unusual morphology that may cause concern for malignancy. Adv Anat Pathol 2010; 17: 91-112.

[6] Travaglino A, Raffone A, Santoro A, Raimondo D, Improda FP, Cariati F, De Marco M, Casadio P, Seracchioli R, Zullo F, Insabato L and Zannoni GF. Risk of recurrence in uterine leiomyoma with bizarre nuclei: a systematic review and meta-analysis. Geburtshilfe Frauenheilkd 2021; 81: 1217-1223.

[7] Ubago JM, Zhang Q, Kim JJ, Kong B and Wei JJ. Two subtypes of atypical leiomyoma: clinical, histologic, and molecular analysis. Am J Surg Pathol 2016; 40: 923-933.

[8] Dastranj Tabrizi A, Ghojazadeh M, Thagizadeh Anvar H, Vahedi A, Naji S, Mostafidi E and Berenjian S. Immunohistochemical profile of uterine leiomyoma with bizarre nuclei; comparison with conventional leiomyoma, smooth muscle tumors of uncertain malignant potential and leiomyosarcoma. Adv Pharm Bull 2015; 5 Suppl 1: 683-687.

[9] Zhang Q, Poropatich K, Ubago J, Xie J, Xu X, Frizzell N, Kim J, Kong B and Wei JJ. Fumarate hydratase mutations and alterations in leiomyoma with bizarre nuclei. Int J Gynecol Pathol 2018; 37: 421-430.

[10] Yavuz E, Güllüoğlu MG, Akbaş N, Tuzlali S, Ilhan R, Iplikçi A and Akhan SE. The values of intratumoral mast cell count and Ki-67 immunoreactivity index in differential diagnosis of uterine smooth muscle neoplasms. Pathol Int 2001; 51: 938-941.

[11] Stojsić Z, Bacetić D, Radević B and Vasiljević JD. A well-differentiated liposarcoma co-existent with leiomyoma. Vojnosanit Pregl 2004; 61: 565-568.

[12] Huffer K, Denley MCS, Oskoui EV and Swartz KJ. Conservation of the cooling agent binding pocket within the TRPM subfamily. Elife 2024; 13: e99643.

[13] Kim HS, Yoon G, Jung YY, Lee YY and Song SY. Fibromyxoid variant of endometrial stromal sarcoma with atypical bizarre nuclei. Int J Clin Exp Pathol 2015; 8: 3316-3321.