

Atypical Uterine Myoma: Difficulties in Diagnosis and Treatment A Case Report

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Abstract:

This article presents a clinical case of surgical treatment of a patient with an atypical uterine myoma located in the lateral pelvic cellular space. The tumor was initially diagnosed as a deep aggressive angiomyxoma based on a transvaginal trephine biopsy. Treatment involved a two-stage approach: preoperative embolization of the tumor vessels followed by radical removal via vaginal access. Final histology with immunohistochemistry (SMA+, h-caldesmon+, CD34-, S100-, HMB45-, Ki67 7%) confirmed the diagnosis of uterine leiomyoma with a rare cord-like growth pattern. This clinical case illustrates the difficulties of differential diagnosis of pelvic tumors, the importance of a multidisciplinary approach to treatment, and the effectiveness of preoperative embolization of tumor vessels.

Keywords: uterine leiomyoma with atypical localization; tumor vessel embolization; myomectomy

Introduction

Uterine leiomyoma is a benign mesenchymal tumor composed of smooth muscle cells, occurring in 20-80% of women of reproductive age. Vaginal localization is extremely rare, accounting for less than 0.1% of cases, with about 300 cases described since Denys de Leyden first observed it in 1733 [1]. Although the etiology of this disease remains a subject of debate, some authors suggest that it arises from remnants of embryonic tissue of blood vessels and smooth muscle fibers [2,3].

Uterine fibroids with atypical locations represent a group of rare retroperitoneal leiomyomas. They are characterized by a distinct clinical presentation that primarily involves impairment of adjacent organ function [4]. Surgical treatment of patients with atypically located tumors is associated with significant surgical trauma, technical complexity, and a high risk of intra- and postoperative complications [5]. The surgical challenges include the complexity of tumor dissection, extensive vascularization, and the risk of injury to the urinary tract organs. Therefore, excision of atypically located myomatous nodules requires thorough preoperative evaluation to identify the specific features of tumor localization, which is key to achieving a successful treatment outcome [6].

Given the extreme rarity of such cases, ultrasound imaging of a mass located near but not connected to the uterus often leads to misdiagnosis as a pelvic

tumor of non-organ origin. In some instances, these fibroids are mistaken for ovarian or rectal tumors. Differential diagnoses for vulvovaginal neoplasms include aggressive angiomyxoma, cellular angiofibroma, angiomyofibroblastoma, lipoma, and fibroma [7]. Computed tomography (CT) scans often provide no additional diagnostic value, whereas magnetic resonance imaging (MRI) is considered more informative. Furthermore, diagnosis should be confirmed by histological examination and immunohistochemistry.

Aim

The aim of this report is to highlight the challenges of differential diagnosis in pelvic tumors, the importance of a multidisciplinary approach to treatment, and the efficacy of preoperative embolization of tumor vessels in reducing intraoperative blood loss.

Presentation

Patient M., 25 years old, was admitted on a scheduled basis to the gynecological department of the Federal State Budgetary Institution “National Medical and Surgical Center named after N.I. Pirogov of the Ministry of Health of the Russian Federation” with complaints of bloody discharge from the genital tract not related to the menstrual cycle. The

medical history indicates that the patient does not visit a gynecologist regularly.

Gynecological status: External genitalia are normally developed, pubic hair growth is of female pattern. The vaginal mucosa is pale pink, with normal relief. A dense, immobile formation measuring 10x6 cm is palpable along the right-side wall of the vagina. The body of the uterus is located above the formation, in anteflexio-versio position, not enlarged, dense, painless on palpation. The appendages on the right and left are not enlarged, painless on examination. Discharge from the genital tract is moderate, mucous.

Ultrasound findings: Uterus measuring 51x47x56 mm. Contours are smooth. Echo structure is heterogeneous. M-echo 9 mm. Right ovary: 33x23x25 mm. The capsule is visible throughout. Echo structure is homogeneous. Follicular apparatus: 5 follicles with a maximum diameter of up to 10 mm. Left ovary: 51x41x36 mm. The capsule is visible throughout. The echo structure is homogeneous. Follicular apparatus: 7 follicles. A formation with a diameter of 40 mm is detected in the stroma with heterogeneous avascular content due to hyperechoic structures and echo suspension. Between the anterior wall of the vagina (possibly connected to it) and the urethra, two solid formations measuring 64x55 mm and 50x46 mm are detected, adjacent to each other. With CDI, blood flow loci are

detected in the structure of the formations and in their capsule. Conclusion: Left ovarian cyst. Solid masses in the pelvis.

Tumor markers: CA-125 - 28.09 U/mL; HE-4 - 43.7 pmol/L; CA 15-3 - 7.95 U/ml; SCC 0.8 ng/ml; AFP 0.762 ng/ml; CA 242 - 10.1 U/ml; CA 19-9 12.1 U/ml; CEA 0.635 ng/ml.

Contrast-enhanced MRI of the pelvis: The uterus is anteflexed, not enlarged: 50x45x53 mm. The cavity is not enlarged, the endometrium is homogeneous. The right ovary measures 32x24x26 mm and contains a moderate number of follicles. The left ovary measures 53x42x38 mm and is enlarged due to two cystic formations measuring 32.5x25x23 mm and 13.5x12.5x11 mm. In the vaginal lumen, on the anterior and right walls, two solid formations with a fairly homogeneous internal structure are determined, with a clear capsule, smooth contours, measuring 51x59x40.5 mm and 42x37x55.5 mm, with pronounced diffusion restriction. The latter formation is closely adjacent to the cervix without signs of infiltration. After the introduction of a contrast agent, its uneven accumulation in the formations is determined. Conclusion: MR image of two large solid formations in the pelvic-perineal region. MR data of focal formations of the left ovary of the endometrioid cyst type (Figs. 1 and 2).

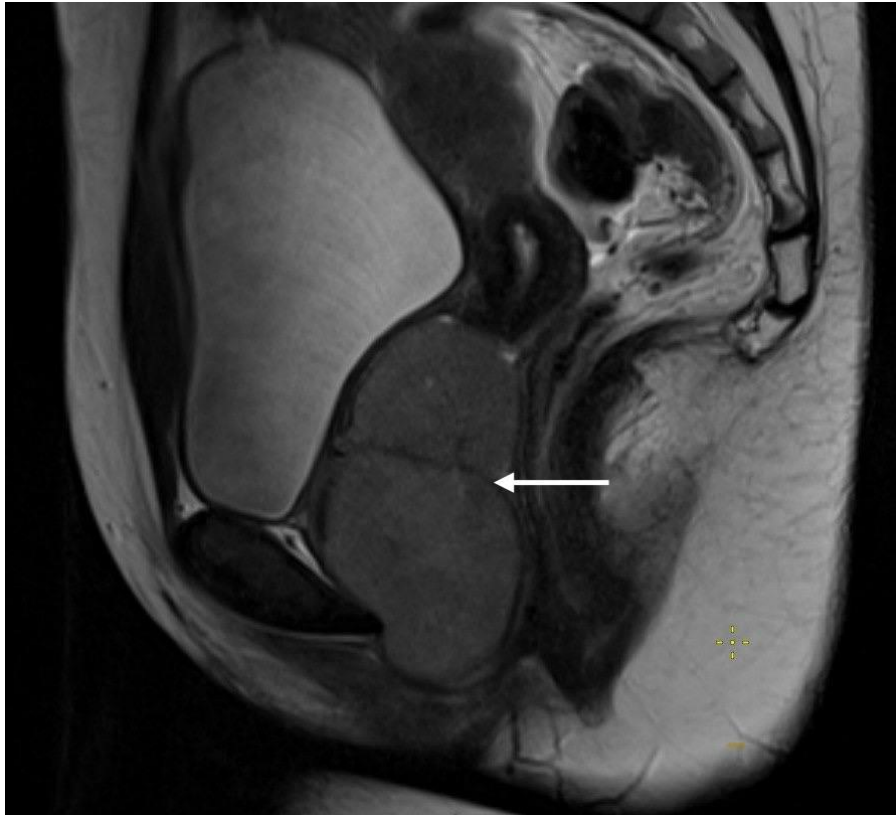


Figure 1: MRI image of a sagittal section of the small pelvis.

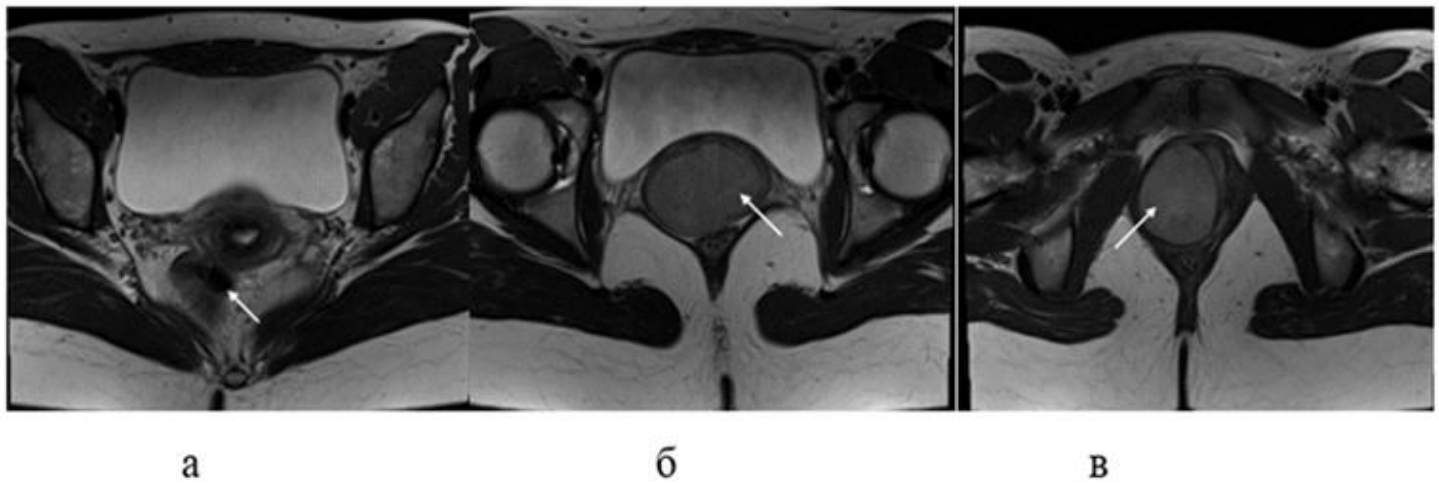


Figure 2: MRI images of axial sections of the small pelvis: a – level of the cervix; b – level of the first formation; c – level of the second formation.

PET-CT findings: Two nodular formations with fairly smooth contours, measuring 59×51 mm and 42×55 mm, with increased metabolism (SUV max 4.9), closely adjacent to the cervix, are detected in the soft tissues of the vaginal and urethral areas. The uterus is not enlarged. The left ovary measures 51×35 mm and contains two cystic formations measuring 32×25 mm and 14×12 mm, while the right ovary measures 22×16 mm. The lymph nodes of the abdominal cavity, retroperitoneal space, and small pelvis are not enlarged. The inguinal lymph nodes are not enlarged. Conclusion: PET-CT signs of nodular formations in the soft tissues of the vaginal area with increased metabolism most likely correspond to a tumor and cysts of the left ovary.

Transvaginal trephine biopsy. Histological examination: Microscopic description – within the limits of the material examined – sections of three small strip-like fragments of the transverse striated muscle with growth of a cellular mesenchymal tumor with a moderately expressed myxoid component, with the presence of vessels of various sizes and configurations, with a few scattered small cells with rounded nuclei, without mitoses and

without foci of necrosis. Pathological-anatomical conclusion: deep (aggressive) angiomyxoma.

Based on instrumental examination methods, a preoperative diagnosis was established: deep (aggressive) angiomyxoma of the pelvic-perineal region. Cyst of the left ovary (presumably endometrioid).

Surgical treatment included sequential endovascular procedures and surgical removal of the tumor.

Stage 1 of surgical treatment: Abdominal aortography. Embolization of pelvic neoplasm vessels.

Under infiltration anesthesia with 1 ml of a 2% lidocaine solution, puncture and catheterization of the left radial artery was performed using a 5F introducer. Heparinization – 2500 IU of sodium heparin was administered intra-arterially (through the introducer). Using a hydrophilic 035 4F 150 cm catheter, sequential catheterization and angiography of the right and left uterine arteries (a. uterina), inferior vesical arteries (a. vesicalis inf.), and internal pudendal arteries (a. pudenda int.) were performed (Figs. 3 and 4).

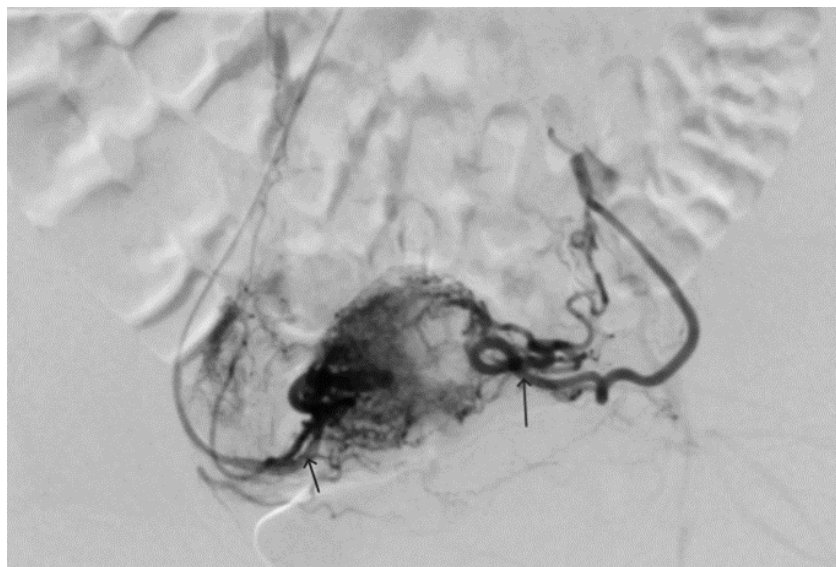


Figure 3: Initial pelvic angiography. The right and left uterine arteries are well contrasted.

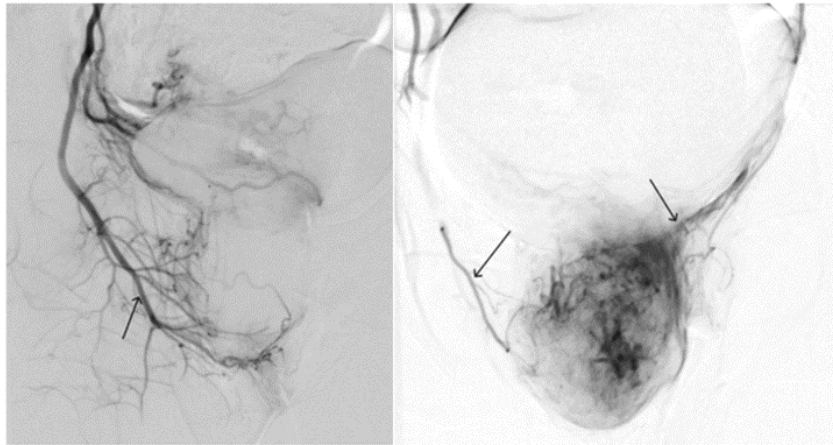


Figure 4: Initial pelvic angiography. Right and left internal genital arteries.

Three syringes of EMBOZENE 900 microns embolization microparticles and two syringes of EMBOZENE 1100 microns were used for embolization. A control angiography was then performed. There was prolonged (more than 10 cardiac cycles) stasis of contrasted blood in the branches of the right and

left uterine, inferior vesical, and internal genital arteries supplying the pelvic organs (Figs. 5 and 6). At the final stage, a pressure bandage was applied to the puncture site of the radial artery for hemostasis. The total fluoroscopy time was 36 minutes.

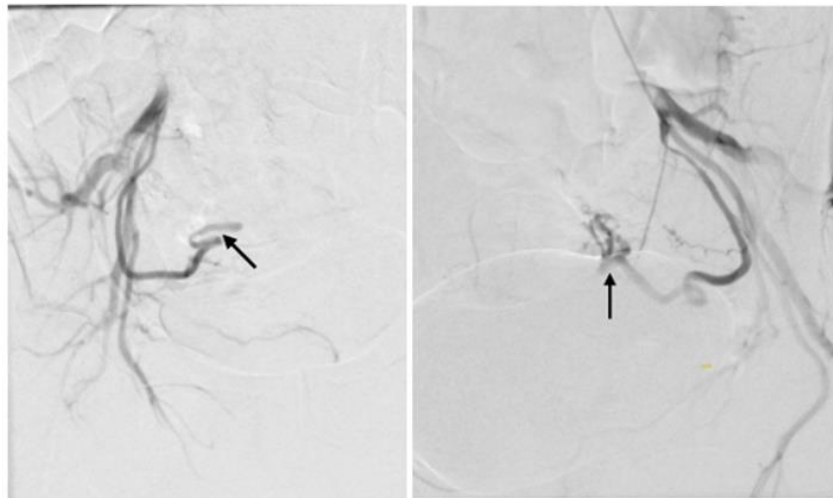


Figure 5: Result of embolization of the right and left uterine arteries.

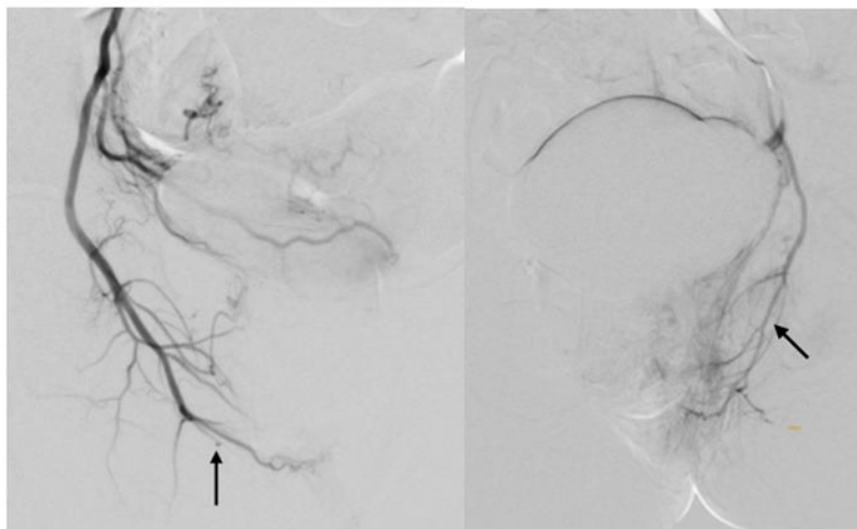


Figure 6: Result of embolization of the right and left internal genital arteries.

Stage 2 of surgical treatment: Transvaginal removal of the pelvic tumor, suturing of the urethral defect. Laparoscopic cystectomy on the left. Coagulation of the endometriosis focus.

Vaginal stage: Under aseptic conditions and endotracheal anesthesia, after treatment of the vagina, an incision was made in the right lateral wall of the vagina in the projection of the pelvic formation (immobile, measuring up to 10x6 cm), located between the urethra, the anterior wall of the vagina, the posterior wall of the bladder, and the pelvic wall. The cervix was not accessible. Next, two formations (6 and 5 cm in diameter) were gradually separated from the pelvic walls and the above-mentioned anatomical structures using blunt dissection. During blunt tissue dissection, a linear defect approximately 1 cm long was revealed in the urethral wall. The neoplasms were removed from the vagina. The defect in the urethra was repaired with separate knotted sutures using 2-0 monofilament thread. Next, the vaginal mucosa was sutured layer by layer with separate sutures using 2-0 monofilament thread. Hemostasis was checked. The vagina was tightly tamponaded. 600 ml of clear urine was drained through the urinary catheter. A permanent urinary catheter was left in place.

Laparoscopic stage: CO₂ pneumoperitoneum was applied in the paraumbilical area. Three trocars were inserted into the abdominal cavity in the paraumbilical, right hypogastric, and suprapubic areas: two 10 mm trocars and one 5 mm trocar. No pathology was detected in the abdominal

cavity. In the small pelvis: The body of the uterus is not enlarged, spherical in shape, marbled in color, with a smooth, shiny serous covering. Left adnexa: The left ovary is enlarged due to a cystic formation with a smooth capsule up to 4 cm in diameter. The left fallopian tube is visually unchanged, the fimbrial section is differentiated. Right adnexa: The right ovary is not enlarged and is visually unchanged. The right fallopian tube is not dilated, visually unchanged, the fimbrial section is differentiated. An endometrioid focus with a diameter of up to 0.2 cm is visualized on the peritoneum of the left lateral canal.

The ovarian tissue above the cyst is opened with an endohook. The cyst capsule is separated with sharp and blunt instruments, opened, and the “chocolate” contents are drained. The endometrioid focus of the pelvic peritoneum is coagulated with an endodissector. Hemostasis control. The specimen was removed from the abdominal cavity through a 10-mm trocar port. Drainage was installed in the pelvic cavity. Gas deflation. Sutures on the incisions of the anterior abdominal wall, aseptic adhesive bandages.

Description of the specimen: 1) two formations of soft-elastic consistency, without signs of decay, 5x5 cm and 6x6 cm in diameter (Fig. 7); 2) capsule of cystic formation measuring 3x2x2 cm.

Duration of surgery: 100 minutes. Blood loss: 600 ml.

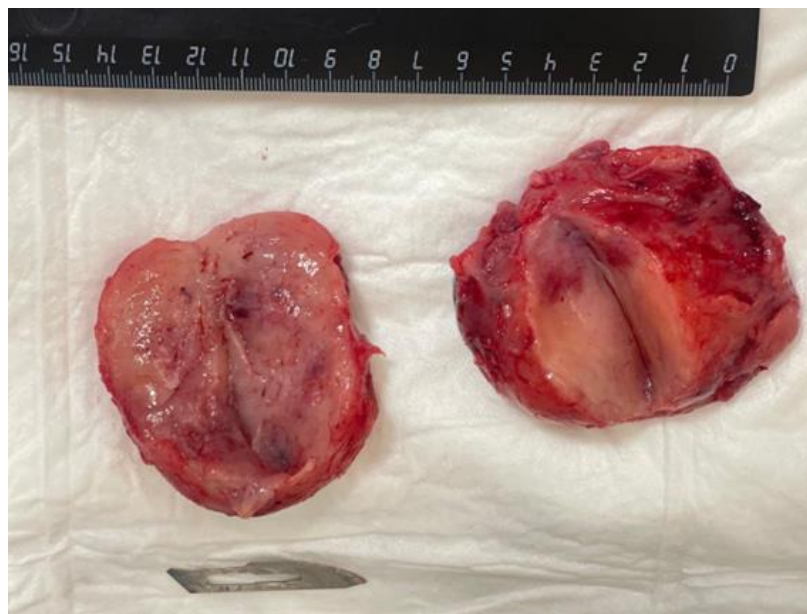


Figure 7: Removed formations of the small pelvis.

Intraoperative diagnosis: Angiomyxoma of the pelvic-perineal region. Endometrioid cyst of the left ovary. External genital endometriosis.

In the early postoperative period, antibacterial therapy, anti-inflammatory, anticoagulant, and symptomatic therapy were administered. The permanent urinary catheter was removed on the 12th day, then a control cystography was performed - there were no signs of leakage in the small pelvis. Postoperative sutures on the anterior abdominal wall were removed on the 10th day. The patient was discharged in satisfactory condition.

Histological conclusion:

Microscopic description: 1) nodular formations are represented by spindle-cell tumors, in some places with an indistinct capsule. There is edema in the

stroma, areas of myxoid type, thin- and thick-walled vessels without extravasates, without necrosis. Spindle cells without atypia, rare mitoses are present. The tumor shows positive staining with antibodies to SMAActin, h-caldesmon. Reaction with antibodies to CD34, S100, and HMB45 antibodies is negative. The Ki-67 proliferation index is 7%.

2) The wall of the ovarian cyst is represented by fibrotic theca, with adjacent areas of cytogenic stroma, siderophages, and hemosiderin deposition. There is no epithelial lining.

Histological conclusion: 1. The morphological picture, taking into account the obtained immunophenotype, corresponds to uterine leiomyoma with cord-like growth. 2. Endometrioid ovarian cyst.

Final diagnosis: Uterine leiomyoma. Endometrioid cyst of the left ovary. External genital endometriosis.

Discussion

The clinical case presented here demonstrates a rare histological variant of uterine leiomyoma—leiomyoma with a cord-like pattern. The exact prevalence of this type of uterine leiomyoma is unknown. This tumor belongs to a group of smooth muscle neoplasms with unusual architecture, which poses significant challenges for morphological verification and requires extensive immunohistochemical examination [8, 9]. In our case, the tumor showed diffuse positive staining for smooth muscle actin (SMA) and h-caldesmon, confirming its smooth muscle origin. The concurrent absence of CD34, S100, and HMB45 expression enabled the exclusion of several differential diagnoses, including schwannoma, angiomyolipoma, perivascular epithelioid cell tumor (PEComa), and melanocytic tumors [10].

Of particular interest is the differential diagnosis between leiomyoma with a cord-like pattern and tumors of the genital tract, such as granulosa cell tumor or Sertoli cell tumor (UTROSCT). Both types of neoplasms may share similar morphological features, including tubular, trabecular, or nested growth patterns [9, 11]. According to published data, the key distinguishing factor lies in the immunophenotype: UTROSCT typically expresses gonadal cord markers (inhibin, calretinin, CD99, Melan-A) with weak or focal expression of smooth muscle markers [11]. In our case, despite the presence of spindle-cell architecture and foci of myxoid changes, the immunohistochemical profile (diffuse expression of SMA and h-caldesmon) allowed the tumor to be classified as a uterine leiomyoma with a cord-like growth pattern [12]. Given the difficulties in histological verification of certain tumors, it is important to recognize that preoperative biopsy results may be inconclusive; however, this does not alter the surgical approach. If such tumors are found to be malignant, the procedure should be regarded as an extended biopsy of the neoplasm.

Over the past 10 years, no more than 50 cases of retroperitoneal tumors with various histological patterns have been described in the global literature. In one case from a clinic in Cameroon, a giant myoma (131.4 × 147.7 mm) was diagnosed, occupying the entire rectovaginal space. The patient underwent a two-stage surgical procedure: diagnostic laparoscopy to assess the degree of displacement of the uterus and adnexa, followed by transvaginal myomectomy with complete removal of the mass and minimal blood loss [13]. In Italy, a case of a 5-cm paraurethral myoma was described, with successful removal via vaginal access and blood loss of less than 100 mL [14].

In addition to this patient, two other patients with tumors located in the lateral pelvic cellular spaces (histological verification - desmoid tumor, angiosarcoma) were operated on in our clinic. In all cases, we performed embolization of the vessels feeding the tumor, which allowed us to avoid massive intraoperative bleeding. This made it possible to create optimal conditions for visualization and work in a “dry” operating field, reducing intraoperative risks.

Conclusion

In this clinical case, the patient achieved a full recovery. The selection of the optimal surgical procedure for patients of reproductive age—myomectomy—enables both radical tumor removal and preservation of reproductive function. The postoperative prognosis is favorable; pregnancy can be planned 3 to 6 months after surgery.

Retroperitoneal pelvic tumors are rare and present with nonspecific clinical manifestations. The presented clinical case illustrates the challenges of differential diagnosis of tumors in this location, underscores the necessity of comprehensive immunohistochemical evaluation, and highlights the technical complexity of surgical interventions, which are best performed in multidisciplinary centers with a collaborative approach.

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