ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2021 August 16; 9(23): 6582-6963

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

Contents Thrice Monthly Volume 9 Number 23 August 16, 2021

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RESPONSIBLE EDITORS FOR THIS ISSUE

NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
ISSN 2307-8960 (online)

LAUNCH DATE
April 16, 2013

FREQUENCY
Thrice Monthly

EDITORS-IN-CHIEF
Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS
https://www.wjgnet.com/2307-8960/editorialboard.htm

PUBLICATION DATE
August 16, 2021

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World J Clin Cases 2021 August 16; 9(23): 6907-6915

DOI: 10.12998/wjcc.v9.i23.6907

ISSN 2307-8960 (online)

CASE REPORT

Uterine tumor resembling an ovarian sex cord tumor: A case report and review of literature

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Author contributions: Zhou FF was responsible for collecting the medical history of the patient and drafting the paper; He YT was responsible for collecting the medical history of the patient; Li Y was responsible for drafting the paper; Zhang M is a pathologist who provided the pathological results; Chen FH reviewed the literature and revised the manuscript; all authors issued final approval for the version to be submitted.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflicting interests.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an

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Abstract

BACKGROUND

Endometrial stromal tumors originate from the endometrial stroma and account for < 2% of all uterine tumors. Uterine tumor resembling an ovarian sex cord tumor (UTROSCT) is a rare histological class of endometrial stromal and related tumors according to the latest World Health Organization classification of female genital tumors. Here, we report a case of UTROSCT in a 51-year-old woman.

CASE SUMMARY

A 51-year-old woman had irregular menses for 6 mo. The patient visited a local hospital for vaginal bleeding. Pelvic computed tomography (CT) showed a mass in the pelvic cavity. Five days later, she came to our hospital for further diagnosis. The results of contrast-enhanced CT and pelvic ultrasound at our hospital suggested a malignant pelvic tumor. She then underwent total removal of the uterus with bilateral salpingectomy. Postoperative histological examination showed that the tumor cells had abundant cytoplasm, ovoid and spindle-shaped nuclei, fine chromatin, a high nucleoplasm ratio, and a lamellar distribution. The findings were consistent with UTROSCT, and the results of immunohistochemical analysis supported that diagnosis. The tumor was International Federation of Gynecology and Obstetrics stage IB. No adjuvant therapy was administered after radical surgery. The patient was followed up for 58 mo, and no recurrence was found.

CONCLUSION

We report a case of UTROSCT with abnormal menstruation as a symptom, which is one of the most common symptoms. In patients with vaginal bleeding, ultrasonography can be used as a screening test because of its convenience, speed, and lack of radiation exposure. For patients with long-term tamoxifen use, routine

Manuscript source: Unsolicited manuscript

Specialty type: Obstetrics and gynecology

Country/Territory of origin: China

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): 0
Grade D (Fair): 0
Grade E (Poor): 0

Received: April 25, 2021
Peer-review started: April 25, 2021
First decision: May 24, 2021
Revised: June 2, 2021
Accepted: June 17, 2021
Article in press: June 17, 2021
Published online: August 16, 2021

P-Reviewer: Jurys T
S-Editor: Ma YJ
L-Editor: Filipodia
P-Editor: Zhang YL

monitoring of the endometrium is recommended. As UTROSCT may have low malignant potential, surgery remains the primary management strategy. Additionally, fertility preservation in patients of childbearing age is a vital consideration.

Key Words: Uterine tumor resembling an ovarian sex cord tumor; Endometrial stromal tumor; Pelvic; Irregular menses; Surgery; Case report

Core Tip: Uterine tumor resembling an ovarian sex cord tumor (UTROSCT) is a rare histological form of endometrial stromal and related tumors. Limited knowledge of the disease can make diagnosis difficult. Here, we present the case of a 51-year-old woman with UTROSCT.

Citation: Zhou FF, He YT, Li Y, Zhang M, Chen FH. Uterine tumor resembling an ovarian sex cord tumor: A case report and review of literature. *World J Clin Cases* 2021; 9(23): 6907-6915
URL: https://www.wjgnet.com/2307-8960/full/v9/i23/6907.htm
DOI: https://dx.doi.org/10.12998/wjcc.v9.i23.6907

INTRODUCTION

Endometrial stromal tumors originate from the endometrial stroma and account for < 2% of all uterine tumors[1]. The latest World Health Organization classification of female genital tumors recognizes uterine tumors resembling an ovarian sex cord tumor (UTROSCT) as a rare histological form of endometrial stromal and related tumors[2]. It was first described by Clement and Scully in 1976[3]. The currently available literature focuses on the diagnosis of UTROSCT and lacks information on the characteristic symptoms and radiological findings. Here, we report a case of UTROSCT in a 51-year-old woman. An attempt was made to review the literature to elucidate the clinical features, treatment options and outcomes of this rare disease to avoid missing or misdiagnosing the disease in clinical practice.

CASE PRESENTATION

Chief complaints

A 51-year-old woman had irregular menses for 6 mo.

History of present illness

The patient had irregular menses for 6 mo. She had pelvic pain and prolonged menstruation but no nausea, vomiting, diarrhea, abdominal bloating, or fever. Pelvic computed tomography (CT) at a local hospital revealed a mass in her pelvis. Five days later, she came to our hospital for further treatment. We investigated whether the patient experienced mild limitations in performing life activities and societal participation, congruent with the domains on the International Classification of Functioning, Disability, and Health.

History of past illness

She had been suffering from hypertension for 2 years, and had been treated with nifedipine extended-release tablets. She underwent surgery to remove a fibroadenoma from her right breast 4 years prior to presentation. The patient had no history of diabetes, heart disease, alcohol consumption, or smoking.

Personal and family history

The patient denied a relevant family history.

Physical examination

Gynecological examination revealed a cervical nodule, approximately 7 cm × 9 cm in size, with a soft texture, no tenderness, a clear boundary, and blood vessel pulsation on the surface without adnexal masses; the vulva, urethra and vagina were normal.

Laboratory examinations

Her blood potassium level was 3.20 mmol/L. All tumor marker results were within the normal range. The reactivity tests for hepatitis B virus, human immunodeficiency virus, syphilis, and hepatitis C virus were all negative.

Imaging examinations

A pelvic ultrasound (US) revealed an 87 mm × 60 mm mass with heterogeneous echo on the right side of the pelvic cavity, consisting of hypoechoic intracystic effusion and a hypoechoic intracystic tumor with local honeycomb changes. Color Doppler imaging showed high degrees of vascularity. The mass had no clear boundary with the cervix and seemed to adhere to it (Figure 1). Pelvic US also suggested multiple uterine fibroids. Pelvic CT revealed a round, low-density mass in the pelvic cavity with uneven internal density. Contrast-enhanced CT showed uneven enhancement in the arterial and venous phases of the scan and no enhancement in low-density areas (Figure 2). The results suggested a malignant pelvic tumor. No enlarged lymph nodes were found in the pelvic cavity.

FINAL DIAGNOSIS

The diagnosis was UTROSCT.

TREATMENT

The patient underwent total removal of the uterus with bilateral salpingectomy. During the surgery, a 100 mm × 80 mm solid mass was detected in the cervix. It had a smooth surface with an intact capsule. The cut surface of the lesion had a solid grayish-yellow appearance, soft cystic areas with hemorrhage and necrosis, and a fish-flesh texture. The intraoperative rapid pathology report suggested a small round cell tumor, and the definite diagnosis was pending routine examination and immunohistochemistry. Postoperative histological examination showed that the tumor cells had abundant cytoplasm, ovoid and spindle-shaped nuclei, fine chromatin, a high nucleoplasm ratio, and a lamellar distribution. These findings were consistent with UTROSCT (Figure 3). Immunohistochemical staining showed that the tumor cells were positive for Ki67 (8% positive), Vim, CD99, CK, and Syn but negative for SMA, ER, PR, myogenin, EMA, HMB45, α-inhibin, CD31, PLAP, CK7, CD56, CD10, WT-1, and caldesmon (Figure 4). No metastasis to the omentum or lymph nodes was observed. To ensure the accuracy of the diagnosis, senior pathologists from other organizations were invited for consultation, and they confirmed the diagnosis of UTROSCT. The International Federation of Gynecology and Obstetrics tumor stage was IB. No adjuvant therapy was administered after radical surgery.

OUTCOME AND FOLLOW-UP

The patient was followed up every 3 mo, and each follow-up examination included a medical history, a physical examination, comprehensive biochemical tests, a chest CT, a vaginal US, and a routine blood examination. An abdominal CT was performed every 6 mo for 58 mo after surgery, and no signs of recurrence or metastasis were detected. The patient's performance of life activities and societal participation had improved.

DISCUSSION

UTROSCT is a rare class of uterine tumors first reported by Clement and Scully in 1976 [3]. They reported 14 cases of UTROSCT, which were divided into two groups based

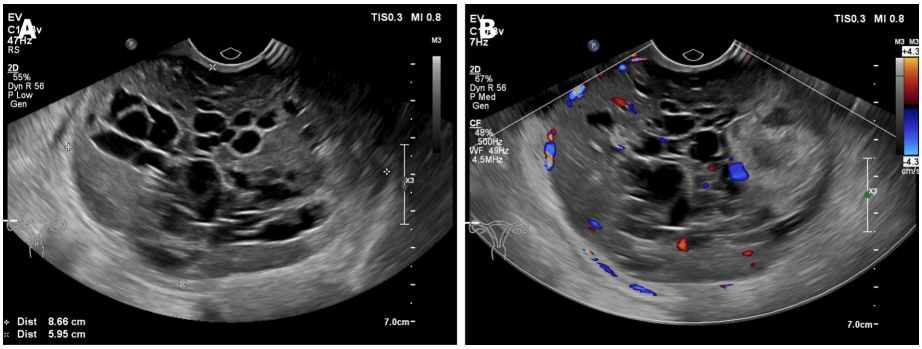

Figure 1 Ultrasound scan. A: An 87 mm × 60 mm mass with a heterogeneous echo is seen on the right side of the pelvic cavity. It consists of a hypoechoic intracystic effusion and a hypoechoic intracystic tumor, with local honeycomb changes; B: Color Doppler imaging shows hypervascularity.

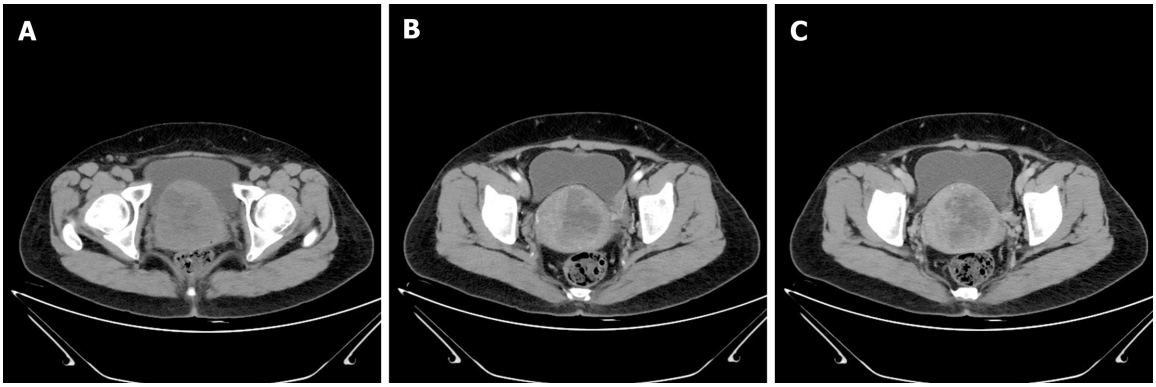

Figure 2 Contrast-enhanced computed tomography. A: A round low-density mass with uneven internal density was present in the pelvic cavity; B, C: Uneven enhancement was present in the arterial and venous phases of the scan and no enhancement in low-density areas.

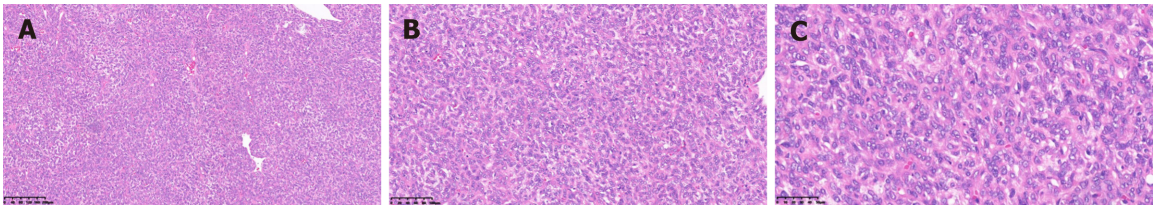

Figure 3 Histopathological findings. A: Hematoxylin-eosin (HE) staining (× 100); B: HE staining (× 200); C: HE staining (× 400).

on the proportion and the appearance of the stromal cell component. Type 1 tumors were endometrial stromal tumors that showed focal epithelial-like differentiation of the type seen in ovarian sex cord tumors, and type 2 tumors—designated UTROSCT—were uterine mural masses with a predominant or exclusive histological appearance of sex cord elements[2]. The origin of UTROSCT is not clear, and the lack of a JAZF1-SUZ12 gene fusion suggests that the origin is the endometrial stroma[4].

To date, including this patient, approximately 90[3-48] cases have been reported in the literature. According to previous reports, the age of onset is 20 to 86 years; the average age is 50.6 years, the median age is 51 years, and the tumor size is 4 to 135 (average 47.6) mm. The patient in this case was a 51-year-old woman, which is consistent with the patient characteristics reported in the literature. Table 1 summarizes the clinical characteristics of the 90 previously reported cases and this case.

In our literature search, we found six patients with UTROSCT with previous or current use of tamoxifen. We therefore suspect that tamoxifen was a causative factor. The effect of tamoxifen on the endometrium has been reported in the literature. Tamoxifen intake can lead to extensive senile cystic atrophy of the human endometrium, leading to endometrial hyperplasia and endometrial polyp formation, and increases the risk of endometrial cancer in postmenopausal women[49]. Unfortunately, published reports on UTROSCT are scarce, and we did not have sufficient data to

Table 1 Summary of the reported characteristics of uterine tumors resembling an ovarian sex-cord tumor

Characteristic		*n* (%)
Age in yr		90
	≤ 30	12 (13.3)
	30-60	47 (52.2)
	≥ 60	31 (34.4)
Location		53
	Uterine wall	35 (66.1)
	Uterine cavity	12 (22.6)
	Uterine wall and cavity	6 (11.3)
Tumor size in mm		72
	≤ 40	36 (50.0)
	40-80	20 (27.8)
	≥ 80	16 (22.2)
Presenting symptom		59
	Postmenopausal bleeding	20 (33.9)
	Abnormal menstruation	20 (33.9)
	Pelvic pain	11 (18.6)
	Elevated prolactin	2 (3.4)
	Incidental	11 (18.6)
Diagnostic modality		30
	US	22 (73.3)
	CT	3 (10.0)
	MRI	5 (16.7)
Accompanied diseases		20
	Leiomyoma	12 (60.0)
	Adenomyosis	4 (20.0)
	Endometrial hyperplasia	2 (10.0)
	Uterine prolapse	2 (10.0)
Surgical approach		75
	TAH+BSO	57 (76.0)
	TAH alone	8 (10.7)
	Mass resection alone	3 (4.0)
	Hysteroscopic mass resection	7 (9.3)
Recurrence/metastasis		52
	Yes	10 (19.2)
	No	42 (80.8)
Status		50
	ANED	44 (88.0)
	AWD	5 (10.0)
	DOD	1 (2.0)

AWD: Alive with disease; ANED: Alive with no evidence of disease; BSO: Bilateral salpingo-oophorectomy; CT: Computed tomography; DOD: Dead of

disease; MRI: Magnetic resonance imaging; TAH: Total abdominal hysterectomy; US: Ultrasound; UTROSCT: Uterine tumor resembling an ovarian sex-cord tumor.

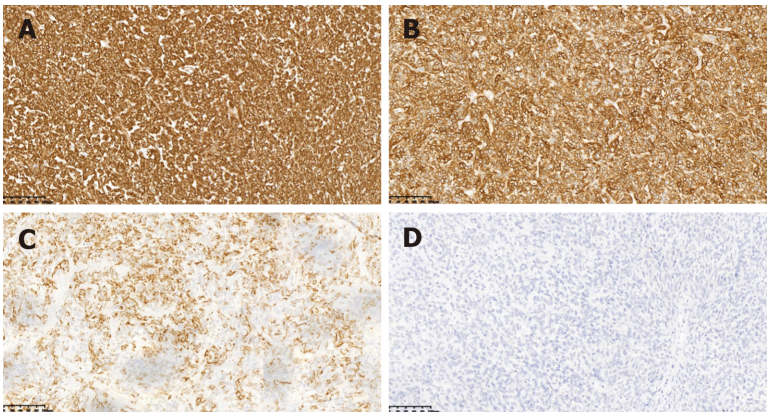

Figure 4 Immunohistochemical staining (× 100). A: Immunostaining for vimentin; B: CD99; C: Cytokeratin; D: α-Inhibin.

draw conclusions.

The diagnosis of UTROSCT is incredibly difficult. The symptoms vary among patients and are not typical in some cases. Therefore, it is easy to miss or misdiagnose the disease. Common symptoms of UTROSCT include postmenopausal bleeding (33.9%)[5], abnormal menstruation, menorrhagia and extended menstruation (33.9%)[14,17], and pelvic pain (18.6%)[19]. We found reports of two patients with UTROSCT who presented with symptoms of hyperprolactinemia, and we found that postmenopausal bleeding and menstrual abnormalities are the most commonly reported symptoms of UTROSCT. The possible mechanism is overgrowth of sex cord elements in endometrial stromal neoplasms or of foci of adenomyosis and endometriosis[50]. The mechanism of hyperprolactinemia remains unclear. However, other reported cases of ectopic hyperprolactinemia with uterine tumors have features in common with this case, and it is possible that they belong to the same tumor category[51].

US, CT, and magnetic resonance imaging (MRI) are useful for detecting UTROSCT[5-11]. Among these techniques, vaginal US is more convenient and less costly for primary diagnosis, while avoiding radiation. According to the literature, 22 (73.3%) cases of UTROSCT were discovered through US. Of the affected patients, ten presented with uterine cavity lesions and five presented with an enlarged uterus. Nine cases included intratumoral cystic components. Of the five reports including MRI, three reported masses described as slightly hyperintense on T2-weighted images and intermediate signal intensity on T1-weighted images.

The diagnosis of UTROSCT is mainly based on the morphological features following hematoxylin-eosin (HE) staining and confirmed by immunohistochemical staining. UTROSCT predominantly has the morphological features of sex cord-like elements wherein tumor cells form cords, trabeculae, tubules, clusters, sheets, and retiform structures. Immunohistochemically, UTROSCT is multiphenotypic, coexpressing smooth muscle markers, cytokeratins, and hormone receptor markers that are usually positive in ovarian sex cord-mesenchymal tumors, including calretinin, inhibin, CD99, CD56, Melan-A, FOXL2, and steroidogenic factor-1 (SF-1)[52,53]. In addition to being positive for calreticulin, the tumors are positive for at least one sex cord marker[54-56]. In the case reported here, the immunohistochemical results showed that the tumor cells were positive for Ki67 (8% positive), Vim, CD99, CK, and Syn; but negative for SMA, ER, PR, myogenin, EMA, HMB45, α-inhibin, CD31, PLAP, CK7, CD56, CD10, WT-1 and caldesmon, consistent with previous published reports.

Standardized treatments for UTROSCT are lacking, perhaps due to its rarity. At present, the preferred treatment method is surgery. The surgical treatment options are either total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH+BCS), TAH alone and mass resection alone. Of the 75 patients in Table 1 who underwent surgery; 57 were treated with TAH+BCS and eight with TAH alone. Hysterectomy is a radical cure, but it is not suitable for patients with fertility requirements. Seven patients with reproductive requirements were treated by hystero-

scopic mass resection. Following treatment, three had spontaneous conception and uncomplicated pregnancies 17 mo, 24 mo, and 13 mo after surgery and all three delivered healthy babies.

Of the 90 patients reported thus far, 52 were followed up for periods of 1 to 384 (mean 56.3) mo; 42 (80.8%) had no recurrence, and 10 (19.2%) had recurrences or distant metastases (lung, abdominal, bladder, mesentery, or pelvic). One patient developed lung and abdominal metastases and died of the disease 9 mo after diagnosis from complications of intra-abdominal tumor spread. In our UTROSCT case, the patient had no fertility requirements, so she underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, and the treatment was effective.

CONCLUSION

This UTROSCT patient presented with abnormal menstruation, which is one of the most common symptoms. In patients with vaginal bleeding, ultrasonography can be used as a screening test because it is convenient, rapid, and lacks radiation involvement. For patients with long-term tamoxifen use, routine monitoring of the endometrium is recommended. As UTROSCT may have a low risk of malignancy, surgery remains the preferred treatment. Preservation of fertility in patients of childbearing age is a key consideration.

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