Recurrent Uterine Tumor Resembling Ovarian Sex Cord Tumor: A Case report and Clinicopathological and Immunohistochemical Analysis

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Case Report

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Abstract

Background: Uterine tumor resembling ovarian sex-cord tumor (UTROSCT) is a considerably rare gynecological tumor which has undetermined pathogenesis but with distinct polyphenotypic immunohistochemical expressions. According to the limited cases and follow-up information in the relevant literature, most of the tumors exhibit indolent or low malignant clinical course and the outcomes of the patients with the tumors generally have a good ending. But for the subset of UTROSCT with aggressive characters, the outcomes of the patients with recurrent neoplasm were not always satisfactory.

Case presentation: In this case report, the recurrence of neoplasm was reported in the pelvic cavity after 53 months of surgery and irregular follow-up. The recurrent neoplasm grew in an invasive manner, and the arrangement of the recurrent neoplasm cells was closer, the nucleus atypia is more pronounced, and the cells demonstrated a more briskly mitotic activity (10 mitotic figures/10 HPF). The Ki67 index increased significantly and the expression of P53 was positive. For the recurrent tumor, both the clinical characteristics and the histological morphology of the recurrent neoplastic cells showed a more malignant behavior. The patient received a palliative resection of pelvic mass and bilateral oophorectomy, and she died of intestinal obstruction caused by the recurrent disease 9 months postoperatively.

Conclusions: UTROSCT should be recognized as a definite malignant potential neoplasm. Based on the clinicopathological, immunohistochemistry parameters and the reviewed previous literature, we speculated that significant mitotic activity and large tumors (≥10 cm) were of value for the aggressive characters of the tumor, and the recurrence of the tumor might lead to a poor prognosis.

Introduction

Uterine tumor resembling ovarian sex-cord tumor (UTROSCT) is a considerably rare gynecological tumor which has undetermined pathogenesis but with distinct polyphenotypic immunohistochemical expressions. Clement and Scully initially narrated the tumors in 1976 and classified those for two subgroups[1]: Type I is an endometrial stromal tumor accompanied by tissues with focally differentiated sex-cord-like areas, and the focal ovarian sex-cord patterns are less than 50% (also called endometrial stromal tumor with sex cord-like elements - ESTSCLE); While type II tumor has a chief or exclusive pattern of ovarian sex-cord cells without recognizable endometrial stroma (also we usually called uterine tumor resembling ovarian sex-cord tumor UTROSCT). In 2021 NCCN Clinical Practice Guidelines for Uterine Neoplasms, UTROSCT is classified as a uterine sarcoma and is described as "bland spindle cell proliferation with extensive sex cord-like differentiation and no endometrial stromal component [2]. To date, only less than 100 cases have been reported in the English-language literature. Due to its rarity and lacking sufficient clinical, pathological, and follow-up information for available cases, the intrinsic molecular mechanisms of the tumors are still unclear. Clinically, ESTSCLE often recurs whereas UTROSCT is usually regarded as indolent or low malignant potential neoplasm [3]. However, several cases of UTROSCT have reported recurrence and metastasis during postoperative follow-up[4-14]. The
The purpose of this article was to provide more information on the recurrence of UTROSCT and was aware of these specific clinicopathological parameters that were of value in predicting malignant behavior for clinicians in dealing with the subset neoplasms with aggressive characters.

**Case Presentation**

In May 2015, a 46-year-old married woman (gravity 5, parity 1) was admitted to the hospital with the complaint of irregular, abnormal vaginal bleeding for 5 months. Sometimes the amount of vaginal bleeding is the same as the usual volume of menstruation, but on the other time it was only a bit. She didn’t present with the complaint of abdominal discomfort. Weight loss was also not reported. Special medical or surgical history was not reported, but a family history of prostate cancer (father) and hypertension (mother) were mentioned. By the pelvic examination, we found a significantly enlarged uterus as the same size uterus of 3 months pregnancy. Transvaginal ultrasonography demonstrated multiple hypoechoic intrauterine masses with distinct border, and the largest one (7.5×7.8×11.9 cm) was located at the fundus of the uterus with a distinct border, and the other two small masses were located at the anterior wall of the uterus (3.9×2.7 cm, 1.2×0.8 cm). The color flow imaging displayed striped blood flow signals around the largest mass (Fig. 1). The thickness of the endometrium was recorded at 1.4 cm and the two ovaries appeared normal. The initial diagnosis was uterine leiomyoma. The result of serum cancer antigen 125 (CA 125), Carcino Embryonic Antigen (CEA), alpha fetal protein (AFP), and human epididymis protein 4 (HE4) was within the normal range. Three months ago, the patient had a diagnostic curettage due to irregular vaginal bleeding for 2 months and the histopathological results were simple hyperplasia at the other hospital. At that time the transvaginal ultrasonography showed the largest size of intrauterine masses was 7.3×7.6×9.7 cm. The patient did not accept medication to adjust the menstrual cycle, and the patient recurred irregular vaginal bleeding more than 2 months ago. Considering the patient’s recurring irregular vaginal bleeding and the enlargement of uterine fibroids, the patient was arranged a laparoscopic total hysterectomy with bilateral salpingectomy. Gross examination displayed an intramural mass of 11 cm in diameter and it had a clear boundary with the surrounding muscles. The section of the mass was yellow, soft, and fleshy. Histologically, the neoplastic cells appeared as round or irregular with eosinophilic, scanty cytoplasm and ovoid nuclei. Mitotic figures were visible (6 mitotic figures per ten high-powered fields, 6 mitotic figures/10 HPF), and necrosis was absent in the background. The uniform neoplastic cells presented anastomosing cords and trabeculae with a reticular architecture and focal interstitial collagenization. There was no muscle infiltration and vascular invasion. Immunohistochemistry (IHC) study revealed diffuse positivity for estrogen receptor (ER), progesterone receptor (PR), and vimentin, weakly positive for CK, and focally and weakly positive for CD99 (Fig. 2). Melan-A, inhibin, Calretinin, Wilm's tumor-1 (WT-1), smooth muscle actin (SMA), Desmin, S-100, D2-40, CD34, CD10, CK5/6, epithelial membrane antigen (EMA), human melanoma black 45 (HMB45) were all negative. The Ki67 proliferative index was about 5%. The other two small mass in the uterus matched the characteristics of fibroids. Then as the results of the morphological features and immunophenotype of the neoplasm cells, the definitive diagnosis was UTROSCT. The patient refused the subsequent
suggestions for radical surgery, chemotherapy, or radiotherapy. The patient then was under clinical and radiological follow-up with vaginal ultrasound every six-twelve months.

In 2019, after 53 months of initial diagnosis and irregular follow-up, the patient presented with complaints of mild and intermittent lower abdominal pain accompanied by frequent urination for three months. Then an abdominal and pelvic mass were found by transvaginal ultrasonography. The PET/CT (positron emission tomography/computed tomography) scan found a giant mass (maximum cross-section was 11.4×10.1 cm) with multiple cystic-solid complexes in the pelvic and abdominal cavity which was closely adherent to the adjacent intestines, bladder, and the top of vagina. FDG (fluorodeoxyglucose)-PET image showed accumulated FDG uptake in the masses (Fig. 3). In the abdominal cavity massive ascites were detected but lymphadenopathy was not found. The serum CA125 level appeared to be slightly increased and reached 37.9 U/mL, the serum CEA, AFP, and HE4 levels were normal. Then the occurrence of a malignant tumor was strongly suspected. But the source of the tumor needed further confirmation. Then a palliative resection of pelvic mass and bilateral oophorectomy were performed. During the operation, the bloody ascites about 3,000 ml were sucked out of the abdominal cavity. Then the yellow and soft mass of about 20×15×10 cm was identified which grew invasively from the pelvic floor fascia to the periphery (Fig. 4). The mass is gravely adhered to the small intestine, colon and the bottom of the bladder. The bilateral ovarian tissues were wrapped in the gap of mass and the intestines. Although the tumor margin was carefully resected, residual lesions that were gravely adhered to the adjacent intestine, bladder, and posterior peritoneum remained. Histologically, both the morphological and IHC aspects of the neoplasm cells were almost identical to those of the previous uterine neoplasm. On microscopic examination, the morphologies of the neoplastic cells were matching with those of the primary uterine neoplasm, and the neoplastic cells consisted of cordlike, and trabecular architectures. But the arrangements of the cells were closer, the nucleus atypia is more pronounced, and the cells demonstrated a more briskly mitotic activity (10 mitotic figures/10 HPF). Immunohistochemical characters were virtually indistinguishable to that of the original uterine neoplasm, but the Ki67 index reached 25% and the expression of P53 was positive. Finally, the diagnosis of recurrence of UTROSCT was confirmed. To further ensure the diagnosis, the pathological results were consulted by senior pathologists from Chinese PLA General Hospital and they verified the diagnosis of UTROSCT. The patient received a cycle of docetaxel and nedaplatin-based chemotherapy. Due to some reasons, the patient did not complete the following chemotherapy. The tumor progressed and she died of intestinal obstruction caused by the recurrent disease after 9 months postoperatively. We obtained a written authorization by the patient's husband to publish the patient's clinical materials of and corresponding images. A written consent is available for inspection by the Editors-in-Chief of this journal.

Discussion

UTROSCT is a specific group of uterine neoplasms with an uncertain histogenesis but with distinct polyphenotypic immunohistochemical expressions. UTROSCT predominantly occur in perimenopausal or menopausal women and there are no specific clinical characteristics for the disease. Most of the patients only present with postmenopausal vaginal bleeding, abnormal menstruation, or pelvic pain[15, 16], with
the image results of an enlarged uterus or a uterine mass similar to a uterine fibroid. The tumors generally exhibit intramural, submucous, and subserous masses with pushing or infiltrative borders. There are no specific imaging characters for the diagnosis of the tumors. It is often difficult to make an accurate diagnosis of UTROSCTs before operation or through intraoperative frozen sections because many benign and malignant lesions show similar histopathologic patterns[17]. Usually, the diagnosis of the disease is an incidental discovery on postoperative histopathological analysis. On macroscopic examination, UTROSCT neoplasms usually have a well-defined or slightly irregular margin, yellow or tan color, with a variably soft to firm consistency. Microscopically, the neoplastic cells of UTROSCT are usually small, round or oval, while the cytoplasm of the cells could be scant, moderate, or abundant[1, 18]. They lay out a variety of patterns that simulating ovarian sex cord tumors, appearing the architectures of anastomosing trabeculae, tubules, cords, nests, and Call-Exner–like bodies[15–17, 19–21]. Necrosis and hemorrhage are unusual in UTROSCT. Several recent studies on the polyphenotypic condition of the tumors speculated that the tumors might derive from ovarian sex cord cells which have been displaced to the uterus during embryogenesis, or pluripotent mesenchymal stem cells[1, 16]. These cells could differentiate into a variety of tissues, and the tumor has a variable IHC profiles with co-expression of epithelial markers (cytokeratin CK, EMA), smooth muscle markers (SMA, Desmin, h-caldesmon), mesenchymal markers (Vimentin), and sex cord markers (α-Inhibin, calretinin, Melan A, CD99, and WT-1) as well as hormonal receptors and miscellaneous markers (ER, PR, CD10, S100)[17, 20].

In the present case, however, the morphology and arrangement of the neoplastic cells conform to UTROSCT, but we found the IHC results of the case were not exactly consistent with previous reports: only one marker for sex-cord tumors was positive (CD99) besides the positive stains of epithelial markers (CK), mesenchymal markers (Vimentin), hormonal receptors (ER and PR). Negative stains for CD10 helped to differentiate from low-grade endometrial stromal sarcoma with sex-cord differentiation[21], negative for HMB-45 was useful to distinguish between UTROSCT and perivascular epithelioid cell neoplasm (PEComa). S100 is usually positively expressed in melanoma or nerve sheath tumors in the uterus and negative stains for it was valuable to rule out these tumors[22]. Krishnamurthy[23] analyzed seven cases and found one or more sex cord markers (α-Inhibin, Melan A, CD99) in addition to variable immunoreactivity for vimentin, estrogen, and progesterone receptors, keratin, actin, and Desmin often strongly suggested a true sex cord differentiation in these tumors. Irving et al[16] concluded that positive expressions for calretinin plus at least 1 of the other three markers (α-Inhibin, Melan A, CD99) might highly reminder the diagnosis of UTROSCT. For this case, the array of architectural patterns for the neoplastic cells is valuable for the diagnosis of UTROSCT. Because of the diagnostic criteria of IHC profiles came from the case reports over the past ten years, we speculate that the results of our case maybe provide a basis for a particular subset of UTROSCT with different pathologic and molecular signatures.

Although the outcomes of the most patients with the tumors generally have a good ending, UTROSCT should be recognized as a definite malignant potential neoplasm because some cases behave with aggressive characters and have the potential of recurrence or extra-uterine spread. To our knowledge, about 21 cases experiencing distant metastasis or recurrence have been reported so far. The metastasis
and recurrent sites included lymph nodes, abdominal and pelvic peritoneum and cavity, lung, bones, ovary, liver, and vaginal vault[4, 8, 15, 24]. The recurrence rate of UTROSCT is not clearly specified in the literature. Moore et al[4] observed eight cases with recurrence in 34 cases and calculated a recurrence rate of 23.5% for UTROSCT. Kavneet et al[11] reported that one out of six cases (16.7%) relapsed within 1 year of diagnosis, while Günsu et al[9] reported a recurrence rate was only 6.3%. The reason for the first high percentage rate attributed to the inclusion of patients with metastases or occurrence in other medical institution referred to their institution for consultation which increased the proportion of relapsed patients in the medical institution. The outcomes of the patients with recurrent neoplasm were not always satisfactory, and the mortality rate was 37.5% (3/8) in the manuscript of Moore[4]. Unfortunately, variations in clinical course and the rarity of the cases make it difficult to identify this subset neoplasm with aggressive characters. It seems that none of the tested immunohistochemistry markers were associated with survival outcome, but clinicopathological parameters are a more credible indicator for the clinical prognosis in these tumors. Hauptmann et al[25] concluded that the histological characteristics including pushing versus infiltrative borders, vascular infiltration, and mitotic activity might indicate an aggressive process of a UTROSCT. Moore et al[4] analyzed the clinical materials of 34 case and concluded that older patients, necrosis, lymphovascular invasion (LVSI), cervical involvement, significant nuclear atypia, and significant mitotic activity often exhibited malignant behaviors with a follow-up from 6 to 135 months. However, only necrosis and significant mitotic activity (≥ 2 mitotic figures/10 HPF) were statistically significant for relapse. Michelle et al[7] reported myometrial invasion, serosal involvement, LVSI, and high mitotic activity were present in these aggressive cases of UTROSCT. All three reports mentioned mitotic activity was a possible predictor of an aggressive course. In a further study of 43 cases, large tumors (≥ 10 cm) were associated with an increased risk of cervical/extra-uterine spread[15]. Our results parallel with these results, and we speculated that 6–8 mitotic figures/10 HPF and large tumors (≥ 10 cm) in the primary tumor were of value for the recurrent. The recurrent tumor grew in an infiltrative manner, and the recurrent cells showed a more closely arrangement, more significant atypia, and the addition of rare pseudoglands. The Ki67 index reached from 5% up to 25%. All these pathological characters indicated a high-grade transformation. Furthermore, we found the positive expressions of P53 in the recurrent tumor cells. P53 immunohistochemistry has been believed as an accurate surrogate reflecting the underlying TP53 mutation status of a tumor which often gives rise to the function in promoting tumorigenesis and a more aggressive tumor profile[26, 27]. Due to the long storage time of the paraffin specimen for the primary tumor, we didn't screen the expression of P53. However, in the recurring tumor, we found that the expression of P53 was positive. We speculate that the role of P53 mutation was not clear in the course of tumor recurrence, but it may be the reason for the increased malignancy of recurring tumors. P53 immunohistochemistry is quite rarely identified in UTROSCT. Among the retrieved articles on UTROSCT, there were 3 articles relevant to the expressions of P53, of which two cases were positive[18, 28] and one case was negative[20]. The two cases with positive stains of P53 both accepted and underwent a hysterectomy with pelvic plus para-aortic lymph node dissection and with no evidence of progressions for a follow-up of 17 months in the second case. It seemed that there was little evidence of the clinical behavior of UTROSCT with P53 mutation. But it might be due to the limited follow-up
period and the definite role of P53 in UTROSCT still requires large numbers of cases and follow-up results.

For the treatment of UTROSCT hysterectomy with or without bilateral salpingo-oophorectomy is typically recommended. However, when the tumor occurs in the reproductive age group, a fertility-preserving protocol of resection of the tumor was also reported[29–31]. Some authors also reported cases for conservative surgical approaches obtaining successful pregnancies and deliveries[32]. But the report emphasized the patients for conservative management should have no risk factors for recurrence and recommend careful follow-up. Miho Sato et al[33] reviewed the cases of UTROSCT with malignant behavior and concluded that a radical surgery including bilateral salpingo-oophorectomy, lymphadenectomy, and omentectomy might lead to a lower recurrence rate than a simple hysterectomy alone. Considering the adverse outcome of this case, we wonder whether giving a second-stage surgery or a radical surgery at that time might change the patient's prognosis.

The limitation of the case report was a lack of molecular analysis. Recently, several series have identified recurring ESR1-NCOA2/3 and GREB1- NCOA1/2 gene fusions in UTROSCT, and recurrences have been documented in a subset of those harboring GREB1 fusions[12, 13, 34]. Instead, we pose that for the recurrent tumor, both the clinical characteristics and the histological morphology of the recurrent neoplastic cells showed a more malignant behavior. In the future, further analyses should be conducted for a better evaluation of UTROSCT with different subtypes, particularly the prognosis, potential treatment, and range of possible molecular events.

**Conclusions**

UTROSCT should be regarded as a definite malignant potential neoplasm. Based on the clinicopathological, immunohistochemistry parameters and the reviewed previous literature, we speculated that significant mitotic activity and large tumors (≥ 10 cm) were of value for the aggressive characters of the tumor. For the subset of UTROSCT with aggressive characters, the recurrence of the tumor might lead to a poor prognosis.

**Declarations**

**Ethics approval and consent to participate**

Written informed consent was obtained from the patient’s husband for the publication of their information and images. A copy of the written consent is available for review by the Editors-in-Chief of this journal. The study was approved by the Research Ethics Committee of Qingdao Central Hospital, the Second Clinical Hospital of Qingdao University.

**Consent for publication**

All authors read and approved the final manuscript before submission.
Availability of data and materials

To be used for all articles, including articles with biological applications.

Competing interests

The authors declare no conflict of interest pertaining to this article.

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Authors’ contributions

Zhao shu-ping: project development and revision of the manuscript.

Wei xiao-qiang: writing and revision of the manuscript.

Tang Meng: writing the manuscript and collection of clinical data.

Liu lili was responsible for pathologic diagnosis and immunohistochemical analysis.

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**Figures**
Figure 1

Transvaginal ultrasonography showed a hypoechoic mass (7.5*7.8*11.9cm) located at the fundus of uterine with distinct border (A). The color flow imaging displayed striped blood flow signals around the mass (B).

Figure 2

Histological and immunohistochemical features of UTROSCT. (A) The neoplastic cells arranged in cords, trabeculae, and solid sheets, with focal interstitial collagenization (hematoxylin and eosin stain, x 200). (B) Mitotic figures were visible (hematoxylin and eosin stain, x 400, arrow). Immunohistochemistry stains showing vimentin diffusely positive (C), CD99 focally and weakly positive (D), ER and PR diffusely positive (E and F), CK weakly positive (G), and the Ki67 proliferative index was about 5% (H).
Figure 3

(A and C) PET'CT scan revealed a giant mass (the maximum cross-section was 11.4* 10.1 cm) with multiple cystic-solid complexes in the pelvic and abdominal cavity which were closely adherent to the adjacent intestines, bladder and the top of vagina. (B and D) FDG (fluorodeoxyglucose)-PET image showed accumulated FDG uptake in the mass.
The yellow and soft mass was identified about $20 \times 15 \times 10$ cm which grew invasively from the pelvic floor fascia to the periphery.
Figure 5

The arrangement of the recurrent neoplastic cells were closer, mitotic figures were 10 mitotic figures/HPF (A, arrow). The Ki67 index reached 25-50% (B) and the expression of P53 was positive (C).

Supplementary Files

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