Submitted: 17 November, 2022 Accepted: 02 March, 2023 Published: 15 October, 2024 DOI:10.22514/ejgo.2024.109

CASE REPORT

European Journal of Gynaecological Oncology

Recurrent uterine tumor resembling ovarian sex cord tumor: clinicopathological and immunohistochemical analysis of a case report and a literature review

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Abstract

Uterine tumor resembling ovarian sex-cord tumor (UTROSCT) is a 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 literatures, 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. This case report reported neoplasm recurrence in the pelvic cavity after 53 months of surgery and irregular follow-up. The recurrent neoplasm grew in an invasive manner. The arrangement of the recurrent neoplasm cells was closer, the nucleus atypia was more pronounced, and the cells demonstrated a more briskly mitotic activity (10 mitotic figures per ten high-powered fields, 10 mitotic figures/10 HPF). The Ki67 index increased significantly. Both the clinical characteristics and 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. UTROSCT with characters for the aggressive process should deserve more attention. Once it relapses, the recurrent tumor of UTROSCT might show a higher malignant tendency and a poor prognosis. Gynecologists and pathologists need to analyze the clinical and histologic assessment in collaboration and better evaluate UTROSCT with aggressive characters.

Keywords

UTROSCT; Recurrence; Aggressive characters; Case report

1. Introduction

Uterine tumor resembling ovarian sex-cord tumor (UTROSCT) is a rare gynecological tumor with undetermined pathogenesis but with distinct polyphenotypic immunohistochemical expressions. In 2023 National Comprehensive Cancer Network (NCCN) Clinical Practice Guidelines for Uterine Neoplasms [1], UTROSCT is listed under the sarcoma category and described as "bland spindle cell proliferation with extensive sex cord-like differentiation and no endometrial stromal component". So far, only more than 200 cases have been reported in English literature. Due to its rarity and lack of sufficient clinical, pathological, and available follow-up information, the intrinsic molecular mechanisms of the tumors are still unclear. Clinically, UTROSCT is generally regarded as indolent or low malignant potential neoplasm [2]. In contrast, several cases of UTROSCT reported recurrence and metastasis during postoperative follow-up [3–13]. The purpose of this paper is to provide more information on the recurrence of UTROSCT and to explore these specific clinicopathological parameters, which are valuable to predict malignant behavior for clinicians when dealing with aggressive subsets of tumors.

2. 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 at some other time it was only a bit. She did not complain abdominal discomfort. No weight loss was reported. Exceptional medical or surgical history were not reported, but a family history mentioned prostate cancer (father) and hypertension (mother). The pelvic examination revealed a significantly enlarged uterus, the same size as the uterus that was pregnant for 3 months. Transvaginal ultrasonography demonstrated that there were multiple hypoechoic intrauterine masses with clear boundary. The largest one (7.5 $\times$ 7.8 $\times$ 11.9 cm) located at the fundus of uteri with

a distinct border, and the other two small masses situated at the anterior wall of the uterus (3.9 × 2.7 cm, 1.2 × 0.8 cm). The color flow imaging displayed the striped blood flow signals around the most prominent mass (Fig. 1). We recorded the endometrial thickness of 1.4 cm, and both ovaries were normal. The primary diagnosis was uterine leiomyoma. The results of serum cancer antigen 125 (CA 125), Carcino Embryonic Antigen (CEA), alpha fetal protein (AFP), and human epididymis protein 4 (HE4) were within the normal range.

In February, the patient had a diagnostic curettage due to irregular vaginal bleeding for 2 months. The histopathological results were simple hyperplasia. Then, the largest intrauterine mass was 7.3 × 7.6 × 9.7 cm by transvaginal ultrasound examination. The patient did not receive medication to adjust the menstrual cycle, and she relapsed irregular vaginal bleeding in March. Therefore, the patient arranged a total laparoscopic hysterectomy with a bilateral salpingectomy.

Gross examination displayed an intramural mass with a diameter of 11 cm, which was clearly defined from the surrounding muscles, but had no capsule. The cross section of the mass was yellow, soft, and fleshy. Histologically, the neoplastic cells were round or irregular. The scanty cytoplasm was eosinophilic, and the nuclei was ovoid. 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 are arrayed into cords and trabeculae with a reticular architecture. There was no muscle infiltration or vascular invasion.

Immunohistochemistry (IHC) study revealed diffusely positivity for estrogen receptor (ER), progesterone receptor (PR), and vimentin, weakly positive for Cytokeratin (CK), and focally and weakly positive for Cluster of Differentiation 99 (CD99). 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% (Fig. 2). The other two small masses 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 of radical surgery, chemotherapy, or radiotherapy. Then, every 6–12 months, the patient was followed up by vaginal ultrasound and radiology.

In October 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 was 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 were closely adhered to the adjacent intestines, bladder, and the top of the vagina. FDG (fluorodeoxyglucose)-PET image showed accumulated FDG uptake in the mass (Fig. 3). In the abdominal cavity, massive ascites was detected, but lymphadenopathy was not found. The serum CA 125 level appeared to be slightly increased and reached 37.9 U/mL (the normal reference range of CA 125 is 0–35 U/mL). The serum CEA, AFP, and HE4 levels were normal. Then the occurrence of a malignant tumor was strongly suspected.

Nevertheless, the source of the tumor needed a further confirmation. Then a palliative resection of pelvic mass and bilateral oophorectomy were performed. During the operation, we sucked out about 3000 mL of bloody ascites from the abdominal cavity. Then a yellow and soft mass of about 20 × 15 × 10 cm was identified (Fig. 4), which grew invasively from the pelvic floor fascia to the periphery. The mass gravely adheres to the small intestine, colon, and the bottom of the bladder. The giant group and the intestines gravely wrapped the bilateral ovarian tissues. Although the tumor margin was carefully resected, residual lesions that were gravely adhered to the surface of adjacent intestine, bladder, and posterior peritoneum remained.

Histologically, both the morphological and IHC aspects of the neoplasm cells were almost similar to those of the previous uterine neoplasm. Microscopically, the neoplastic cells' morphologies matched with those of the primary uterine

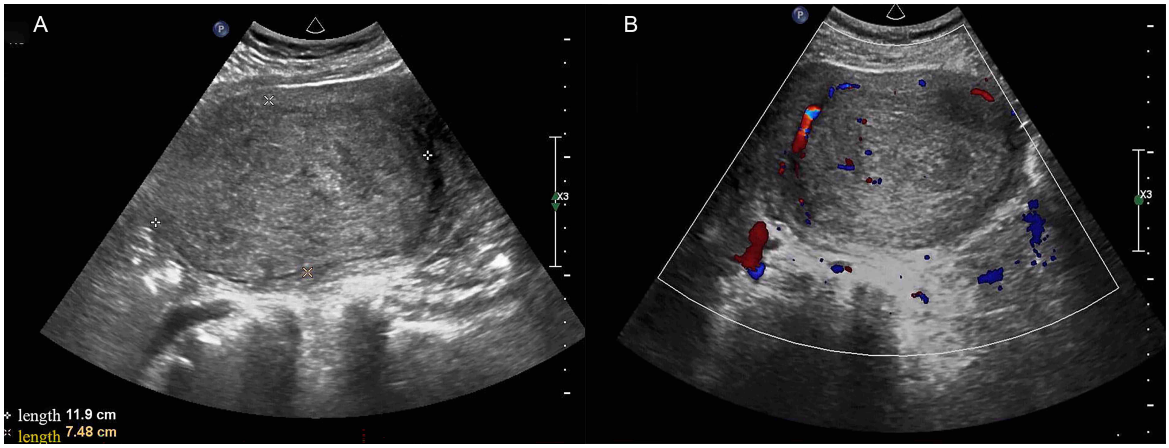

FIGURE 1. Images of ultrasonic examination of UTROSCT. (A) Transvaginal ultrasonography showed a hypoechoic mass (7.5 × 7.8 × 11.9 cm) located at the fundus of uterine with distinct border. (B) The color flow imaging displayed striped blood flow signals around the mass.

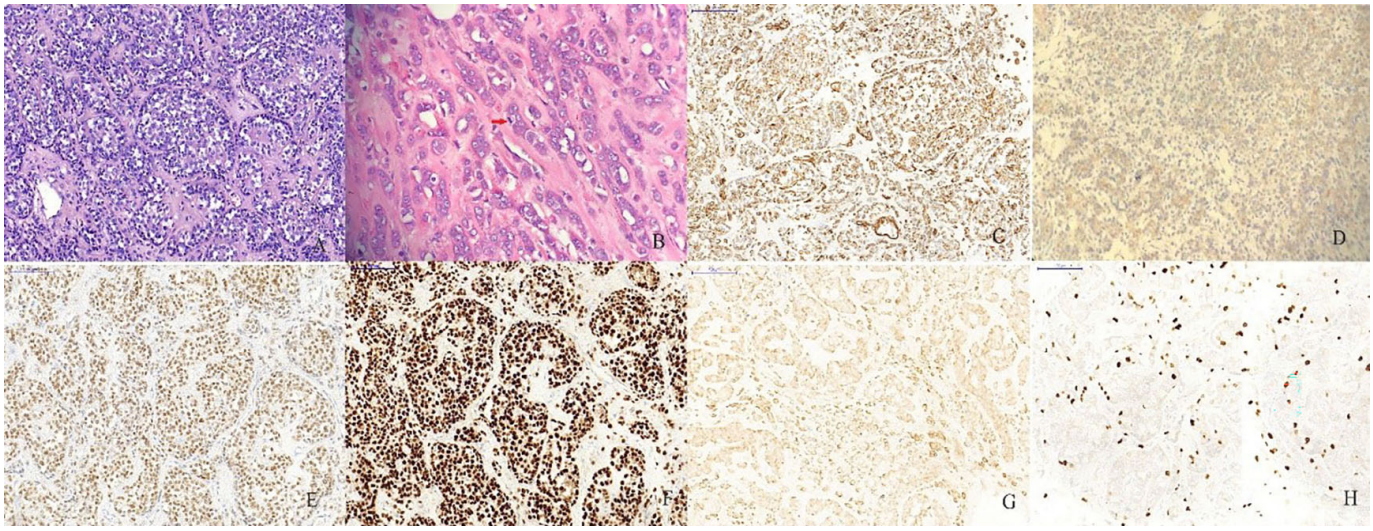

FIGURE 2. Histological and immunohistochemistrical features of UTROSCT. (A) The neoplastic cells arranged in cords, trabeculae, and solid sheets, with focal interstitial collagenization (hematoxylin and eosin stain, ×200). (B) Mitotic figures were visible (hematoxylin and eosin stain, ×400, arrow). (C) Immunohistochemistry stains showing vimentin diffusely positive. (D) CD99 focally and weakly positive. (E,F) ER and PR diffusely positive. (G) CK weakly positive. (H) and the Ki67 proliferative index was about 5%.

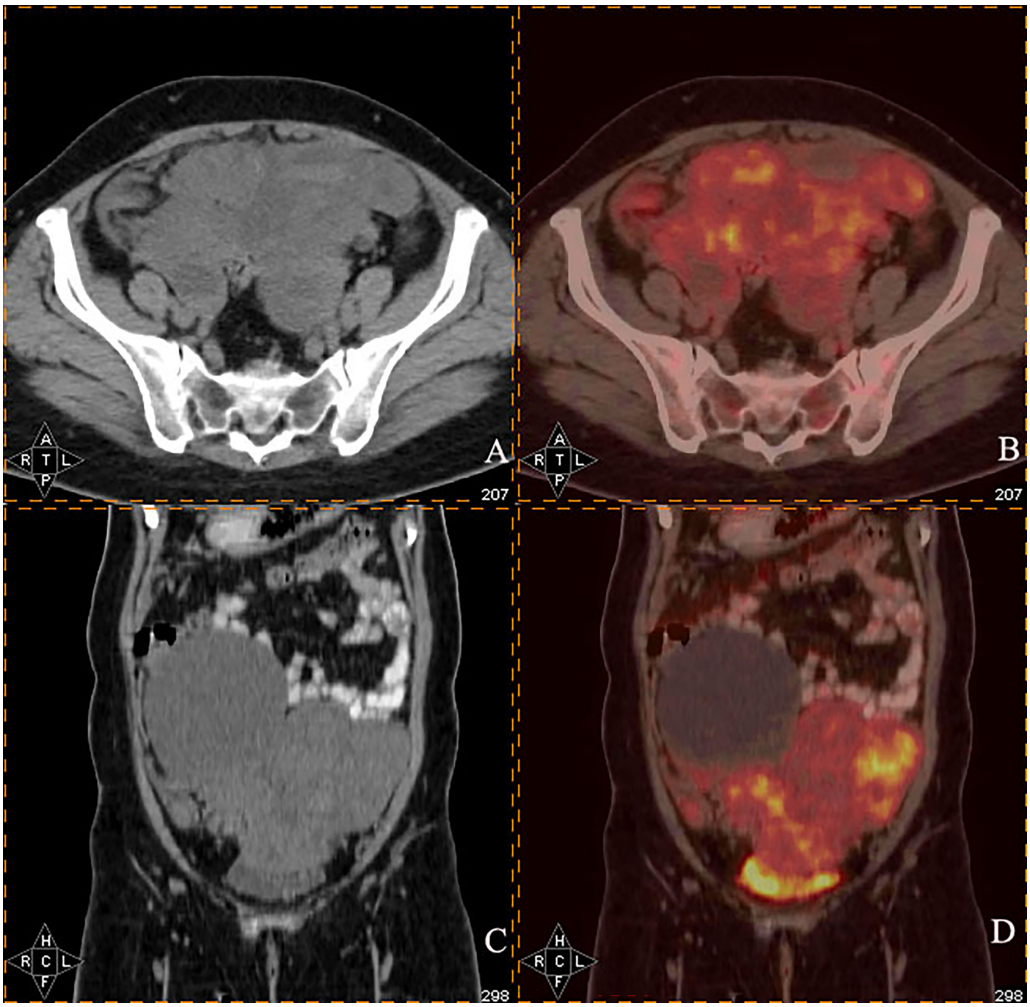

FIGURE 3. PET-CT images of the UTROSCT patients after recurrence. (A,C) PET/CT scan revealed a giant mass (the maximum cross-section was 11.4 × 10.1 cm) with multiple cystic-solid complexes in the pelvic and abdominal cavity which were closely adherent to the adjacent intestines, bladder and the top of vagina. (B,D) FDG (fluorodeoxyglucose)-PET image showed accumulated FDG uptake in the mass.

FIGURE 4. Resected the recurrent tumor tissue. The yellow and soft mass was identified about 20 × 15 × 10 cm which grew invasively from the pelvic floor fascia to the periphery.

neoplasm, and the neoplastic cells consisted of cord-like, and trabecular architectures. However, the arrangements of the cells were closer, the nucleus atypia was more pronounced, the cells demonstrated a more brisk mitotic activity (10 mitotic figures/10 HPF), and necrosis could be found in some areas of the background. Immunohistochemical characters were virtually indistinguishable from the primary uterine neoplasm, but the Ki67 index reached 25%, and a wild type p53 expression was detected (Fig. 5). Finally, the diagnosis of UTROSCT recurrence was confirmed. To further ensure the diagnosis, the senior pathologists in General Hospital of the Chinese People's Liberation Army consulted the pathological results, and they verified the diagnosis of UTROSCT. The patient received a cycle of docetaxel and nedaplatin-based chemotherapy. For 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.

3. Discussion

UTROSCT is a specific group of uterine neoplasms with an uncertain histogenesis but with distinct polyphenotypic immunohistochemical expressions. UTROSCT predominantly occurs in perimenopausal or menopausal women, and there are no specific clinical characteristics for the disease. Most patients only present with postmenopausal vaginal bleeding, abnormal menstruation, or pelvic pain [14, 15], 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. Because UTROSCT often shows a similar histopathologic pattern with many benign and malignant lesions, it is often difficult to accurately diagnose UTROSCT before an operation or through intraoperative frozen sections [16]. Usually, the diagnosis of the disease is an incidental discovery based on postoperative histopathological analysis. On macroscopic examination, UTROSCT neoplasms generally have a well-defined or slightly irregular margin, yellow or tan color, with a variably soft to a 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 [17]. They layout a variety of patterns that simulate ovarian sex cord tumors, appear the architectures of trabeculae, tubules, cords, nests, and Call-Exner—like bodies [14–16, 18–20]. Necrosis and hemorrhage are unusual in UTROSCT. On the polyphenotypic condition, several recent studies speculate that the tumors might derivate from pluripotent mesenchymal stem cells, or the ovarian sex cord cells, which have displaced to the uterus during embryogenesis [15]. These cells could differentiate into a variety of tissues. The tumor has 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) [16, 19].

In the present case, the morphology and arrangement of the neoplastic cells conform to UTROSCT. However, the IHC results of the case were not precisely consistent with previous reports. We only saw 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 differentiate from low-grade endometrial stromal sarcoma with sex-cord differentiation [20], negative for HMB45 helped 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 were valuable to rule out these tumors [21]. Krishnamurthy [22] 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.* [15] 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. In this case, the array of architectural patterns for the neoplastic cells is valuable for diagnosing UTROSCT.

Although most patients with UTROSCT generally have a good ending, some patients should deserve enough attention because some cases behave with aggressive characters and have the potential of recurrence or extra-uterine spread. To our knowledge, about 21 instances 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 [3, 5, 7, 14, 23]. The recurrence rate of UTROSCT is not specified in the literature. Moore *et al.* [3] observed eight cases with recurrence in 34 patients and calculated a recurrence rate of 23.5% for UTROSCT. Kavneet *et al.* [10] reported that one out of six cases (16.7%) relapsed within 1 year of diagnosis, while Günsu *et al.* [8] announced a recurrence rate of only 6.3%. The first high percentage rate benefited

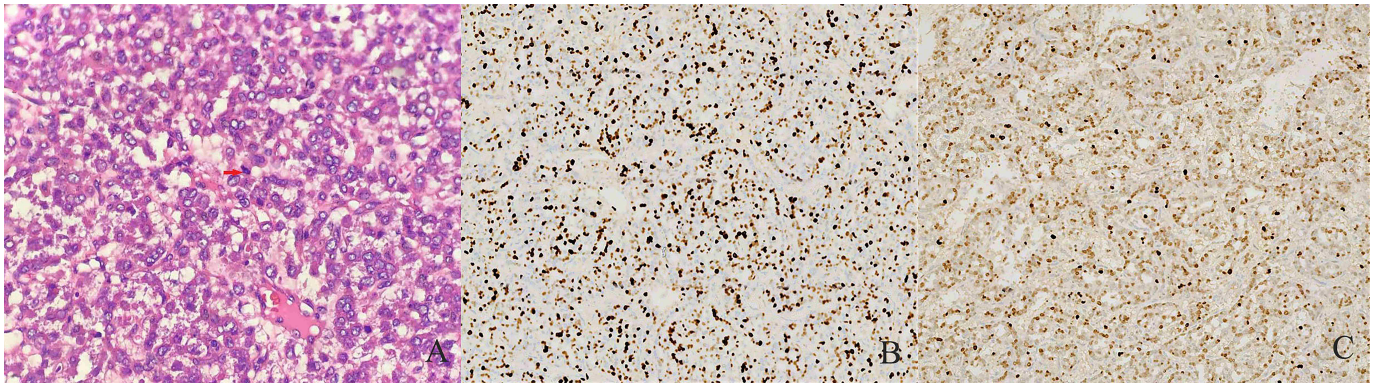

FIGURE 5. Histological and immunohistochemistrical features of recurrent UTROSCT. (A)The arrangement of the recurrent neoplastic cells were closer, mitotic figures were 10 mitotic figures/HPF (arrow). (B) The Ki67 proliferative index reached 25%. (C) The expression of wild type p53 was positive.

from the consultative patients with metastases or occurrences coming from other medical institutions, 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 [3]. Unfortunately, variations in clinical course and the rarity of the cases make it challenging to identify this subset neoplasm with aggressive characters. It seems that none of the tested immunohistochemistry markers were associated with survival outcomes, but clinicopathological parameters are a more credible indicator for the clinical prognosis of these tumors. Hauptmann *et al.* [24] 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.* [3] analyzed the clinical materials of 34 cases 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 ($\geq$2 mitotic figures/10 HPF) were statistically significant for relapse. Michelle *et al.* [6] reported that myometrial invasion, serosal involvement, LVSI, and high mitotic activity were present in these aggressive cases of UTROSCT. All three reports mentioned that mitotic activity was a possible predictor of an aggressive course. In a further study of 43 cases, large tumors ($\geq$10 cm) were associated with an increased risk of cervical/extra-uterine spread [14]. Accordingly, in the present case, the significant mitotic activity (6 mitotic figures/10 HPF) and large tumors might indicate a poor prognosis. In the recurrent tumor, it grew in an infiltrative manner, the nucleus of the recurrent cells showed more significant atypia, and the cells demonstrated a more briskly mitotic activity. The Ki67 index reached from 5% up to 25%. All these pathological characters indicated a high-grade transformation. We found a wild type p53 expression in the recurrent tumor cells. Wild type p53 (a tumor suppressor protein) is a sequence-specific transcription factor that could be activated by genotoxic stress, leading to cell cycle arrest and DNA repair, or inducing apoptosis in damaged cells [25]. 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 sparsely positive [17, 26] and one case was negative [19]. The value of p53 expression for the pathogenesis or the prognostic value of UTROSCT might need more cases.

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 resecting the tumor was also reported [27–29]. Some authors also reported cases with conservative surgical approaches obtaining successful pregnancies and deliveries [30]. Nevertheless, the report emphasized that patients for conservative management should have no risk factors for recurrence and recommended careful follow-up. Miho Sato *et al.* [31] 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, there is an increasing reliance on ancillary genetic/molecular analysis in clinical practice to classify uterine sarcomas and predict prognosis accurately. Several series have identified ESR1-NCOA2/3 (Estrogen Receptor 1-Nuclear Receptor Coactivator 2/3) and GREB1-NCOA1/2 (Growth Regulation by Estrogen in Breast Cancer 1-Nuclear Receptor Coactivator 1/2) gene fusions in UTROSCT. Furthermore, UTROSCT with GREB1-rearrangement tended to be larger and more mitotically active, displayed prominently trabecular or cord-like arranged tumor cells, and often inconspicuous sex-cord differentiation. The tumor group appears to behave more aggressively than ESR1-rearranged UTROSCT [11, 12, 32]. Similarly, the cells in our primary tumor were arranged into a reticular architecture with anastomosing cords and trabeculae. The results of IHC showed that only one marker for sex-cord tumors was positive (CD99), but negative for relatively specific sex-cord markers (α-inhibin, calretinin, FOXL2 (Forkhead box L2) and SF-1 (Steroidogenic Factor-1). On the other hand, the primary diameter was 11 cm, and signif-

icant mitotic activity (6 mitotic figures/10 HPF) seems to support the more aggressive subtype of UTROSCT with GREB1-rearrangement. Therefore, we speculate that UTROSCT might deserve separate consideration for two groups, despite the overlapping clinical and histopathological features and belonging to the same disease spectrum. A comprehensive molecular analysis such as RNA-sequencing is not routinely performed or accessible by cost consideration in many pathological laboratories. In the future, gynecologists and pathologists in clinical practice need to analyze the clinical and histologic assessment in collaboration and better evaluate UTROSCT with different subtypes, particularly the prognosis, potential treatment, and range of possible molecular events.

4. Conclusions

Based on the clinicopathological, immunohistochemistry parameters and the reviewed previous literature, we speculated that UTROSCT with characters for the aggressive process should deserve more attention. Once it relapses, the recurrent tumor of UTROSCT might show a more malignant behavior and a poor prognosis. Gynecologists and pathologists need to analyze the clinical and histologic assessment in collaboration and better evaluate UTROSCT with aggressive characters.

AVAILABILITY OF DATA AND MATERIALS

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

AUTHOR CONTRIBUTIONS

SPZ—project development and revision of the manuscript. XQW—writing and revision of the manuscript. MT—writing the manuscript and collection of clinical data. BXM—writing the manuscript and collection of clinical data. LLL—was responsible for pathologic diagnosis and immunohistochemical analysis. All authors read and approved the final manuscript before submission.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

We obtained a written authorization from the patient's husband to publish the patient's clinical materials and corresponding images. Written consent is available for inspection 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 (IEC-AF-033-04.2).

ACKNOWLEDGMENT

The authors thank Zhao peng (Biotherapy Center of Qingdao Central Hospital) for the critical reading of this manuscript. I would like to express my gratitude to all those who helped us during the writing of this manuscript.

FUNDING

This research received no external funding.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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How to cite this article: Xiao-qiang Wei, Meng Tang, Ben-xu Ma, Li-li Liu, Shu-ping Zhao. Recurrent uterine tumor resembling ovarian sex cord tumor: clinicopathological and immunohistochemical analysis of a case report and a literature review. European Journal of Gynaecological Oncology. 2024; 45(5): 197-203. doi: 10.22514/ejgo.2024.109.