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Recurrence in Uterine Tumors with Ovarian Sex-Cord Tumor Resemblance: A Case Report and Systematic Review

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ABSTRACT

Objective: The aim of this study was to evaluate the prognostic factors of recurrence in uterine tumors resembling ovarian sex-cord tumors (UTROSCT) and to determine clinical-pathological characteristics, treatment options and outcome.

Material and Method: An electronic literature search was conducted from 1976 to 2018. After the comprehensive evaluation and conjunction with our case, the study included 79 cases.

Results: The median age at initial diagnosis was 49 years (range; 16-86 years). The age was under 40 years in 21 (26.6%) patients. Whereas 68 patients underwent at least hysterectomy, 9 patients had organ sparing surgery. There was necrosis in 4 (5.1%) patients, atypia in 16 (20.3%) patients, and infiltrative tumor border in 34 (43%) patients. At least one mitosis per 10 high power fields was determined in 36 (45.5%) patients. The tumor involved at least part of the myometrium in 54 (68.3%) patients. Median follow-up time was 30 months (range; 3-296 months). Recurrence was determined in 5 (6.3%) patients. The disease free survival (DFS) was significantly related only to surgery type. None of the pathologic features were associated with DFS. The 5-year DFS was 86% and 96% in patients who underwent organ sparing surgery or not, respectively (p=0.038).

Conclusion: The accurate pathologic diagnosis of UTROSCT has great value in shaping surgical management and management during the follow-up period. Organ sparing surgery was related to poor DFS. Although recurrence is rare, it should be kept in mind for patients with UTROSCT.

Key Words: Uterine neoplasms, Recurrence, UTROSCT, Surgery, Prognosis

INTRODUCTION

Uterine tumors with sex-cord-like elements can be divided into two groups. The first group of tumors is called endometrial stromal tumors with sex cord-like elements (ESTSCLEs) involving endometrial stromal neoplasms with focal areas (<50%) resembling ovarian sex-cord elements. The second group of tumors is called uterine tumors resembling ovarian sex-cord tumors (UTROSCTs) corresponding to uterine tumors with a predominant or exclusive pattern similar to ovarian sex-cord tumors (1, 2). This morphologic differentiation is clinically significant because these tumors have different biological behaviors. ESTSCLE has a tendency for recurrence and metastases, whereas UTROSCT usually shows more benign clinical behavior (3). UTROSCTs rarely recur and are thus considered uterine tumors with low malignant potential (4). UTROSCTs are extremely rare tumors mostly documented as case series in the literature; therefore, it is difficult to draw a distinct conclusion about the management or recurrence rates of these tumors.

In this study, a recurrent case of UTROSCT is presented. This analysis evaluated prognostic factors related to UTROSCT recurrence and determined clinical-pathological characteristics, treatment options, and outcomes of UTROSCT.

CASE REPORT

A 61-year-old patient was referred to our gynecologic-oncology clinic because of a UTROSCT diagnosis. She underwent total abdominal hysterectomy (TAH) and bilateral salpingo-oophorectomy (BSO) with a diagnosed pelvic mass at another center. The largest diameter of her tumor was 70 mm.

In the differential diagnosis of UTROSCT, we considered ESTSCLE, low-grade endometrial stromal sarcoma, epithelioid leiomyoma, and endometrioid carcinoma

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with sex-cord-like elements. Microscopic evaluation of the specimen was reviewed by pathologists with expertise in gynecologic oncology. The pathologic result showed an endometrial-located tumor. There was a suspicion of superficial focal myometrial invasion. Microscopic evaluation showed anastomosing cords, trabeculae, nests, and tubules of epithelial-like cells found in a fibroblastic stroma (Figure 1). The stromal cells had mild pleomorphism and no necrosis. The mitotic index was 2 per 10 high power fields (HPF). The neoplasm had an infiltrative growth pattern, but there was no vascular invasion. Immunohistochemical analysis was performed. The neoplasm was positive for calretinin, vimentin (Figure 2) CD56 (Figure 3), estrogen receptors (Figure 4), progesterone receptors, Wilms' tumor Protein 1 (Figure 5), synaptophysin, and chromogranin. However, the neoplasm was negative for CD10, EMA, desmin, CD99, and inhibin.

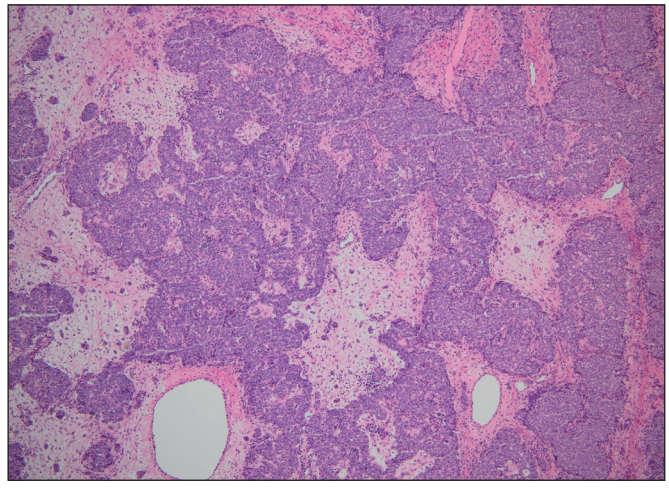

Figure 1: Anastomosing cords, trabeculae, nests and tubules of epithelial-like cells were lying in a fibroblastic stroma. The stromal cells had mild pleomorphism and no necrosis (H&E; x 100).

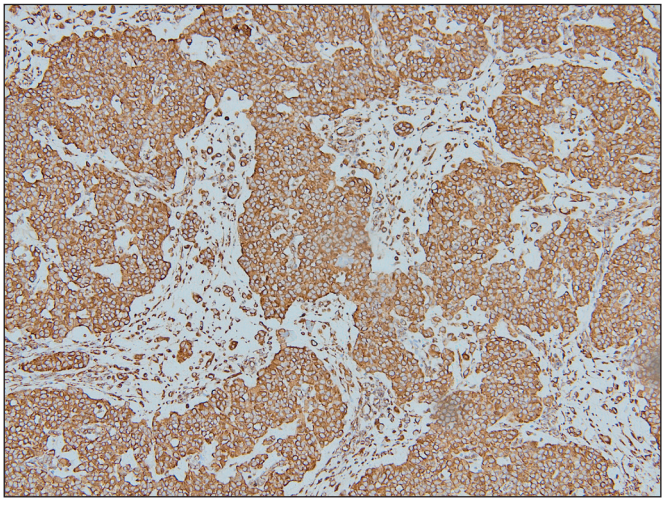

Figure 2: Vimentin positivity (IHC; x100).

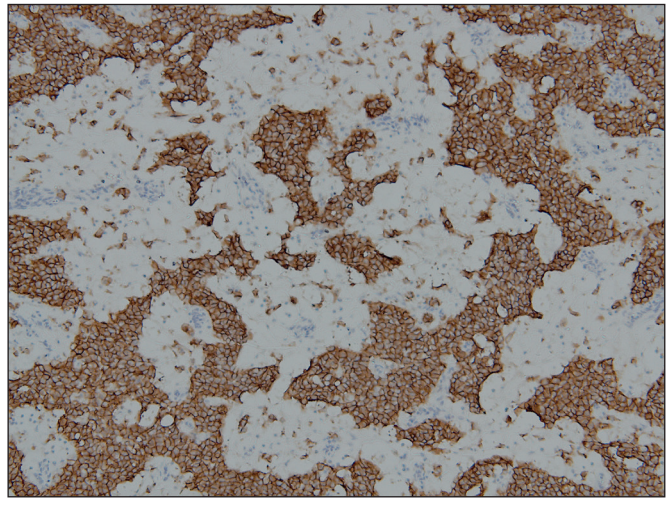

Figure 3: CD56 positivity (IHC; x100).

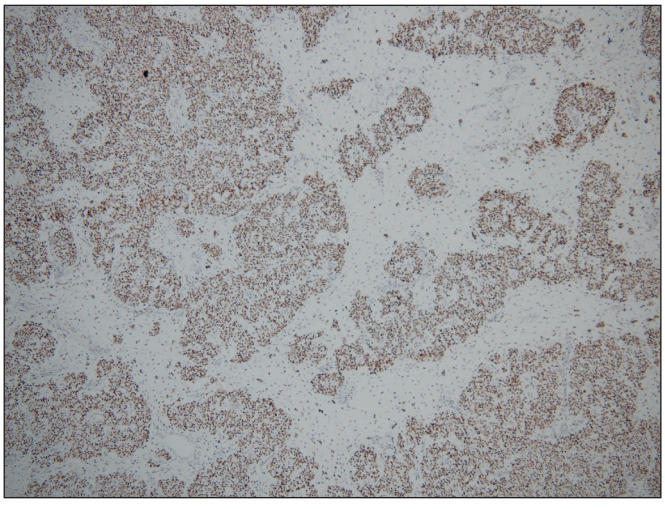

Figure 4: Estrogen receptor positivity (IHC; x100).

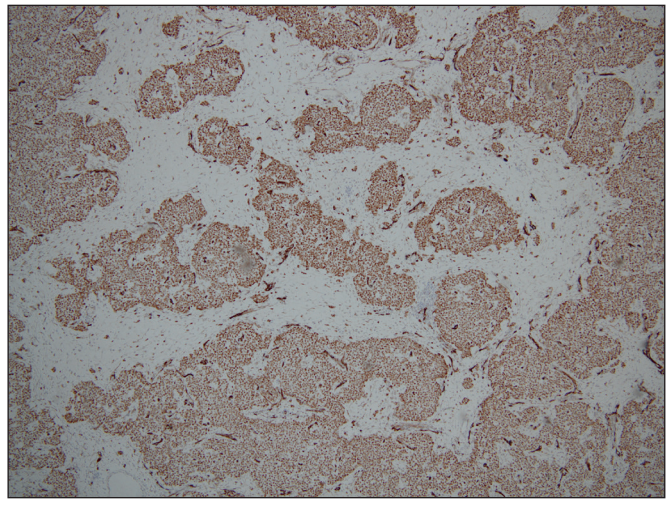

Figure 5: Willms' tumor protein-1 (WT1) positivity (IHC; x100).

According to these findings, the pathologic diagnosis was reported as UTROSCT. A 50 mm pelvic mass was determined incidentally via ultrasonography and magnetic resonance imaging during follow-up at 60 months after the initial diagnosis.

Upon diagnosis, the patient underwent cytoreductive surgery with pelvic mass excision, omentectomy and a tumoral implant resection from the spleen; no tumor was visible at the completion of the surgery. The pathology result of the recurrent disease was reported as UTROSCT with pathological features similar to the initial tumor. The patient received adjuvant hormone therapy for one year, first with megestrol acetate (ceased because of the side effects) and then with letrozole. Three months after completing therapy, she underwent cytoreductive surgery because of secondary recurrence in the pelvis and abdomen. There was a pelvic mass behind the bladder and tumoral implants on the anterior surface of the abdominal wall. At the end of the surgery, maximal cytoreduction with no visible tumor was achieved. According to the medical oncologist's suggestion, adjuvant chemotherapy was planned, but the patient refused chemotherapy. She is alive and disease free seven months after secondary surgery. The follow-up time was 83 months from initial diagnosis.

MATERIALS and METHODS

Literature Review

A systematic review of the medical literature was performed to identify articles about uterine tumors resembling ovarian sex cord tumors. The electronic literature search was conducted from 1976 to January 2018 using PubMed/MEDLINE for English language abstracts. The search included the following medical subject headings (MeSH) or keywords: 'uterine tumor resembling ovarian sex cord tumors' and 'UTROSCT'. After the search was completed, 63 articles were found. After the first evaluation, 23 articles were excluded because of the detailed reasons in the research chart (Figure 6). After a comprehensive evaluation of all cases (N=120) from 40 articles (2, 5-42), the study excluded ESTSCLE cases (n=14) and patients who only underwent curettage without detailed clinical data such as therapy type after curettage, follow-up time, and recurrence status (n=3). Additionally, cases that were referred as consultations (n=32) or not clarified (n=2) as which was the institutional were not included in the study to avoid confusion related to probable case duplication (43). The study evaluated a total of 79 cases.

Tumor size was defined as the largest tumor diameter in the uterus. Tumors were categorized into three types: endometrial (polypoid and confined to the endometrium), myometrial (involving at least part of the myometrium but no serosa; this included submucosal tumors on hysteroscopy), and serosal (involving all the uterine layers, including the serosa). The presence of mitosis was accepted as the presence of at least 1 mitosis per 10 HPF. Surgery types were categorized as either organ-preserving (including myomectomy, hysteroscopy, uterine mass excision, and transvaginal mass extirpation) or non-organ-

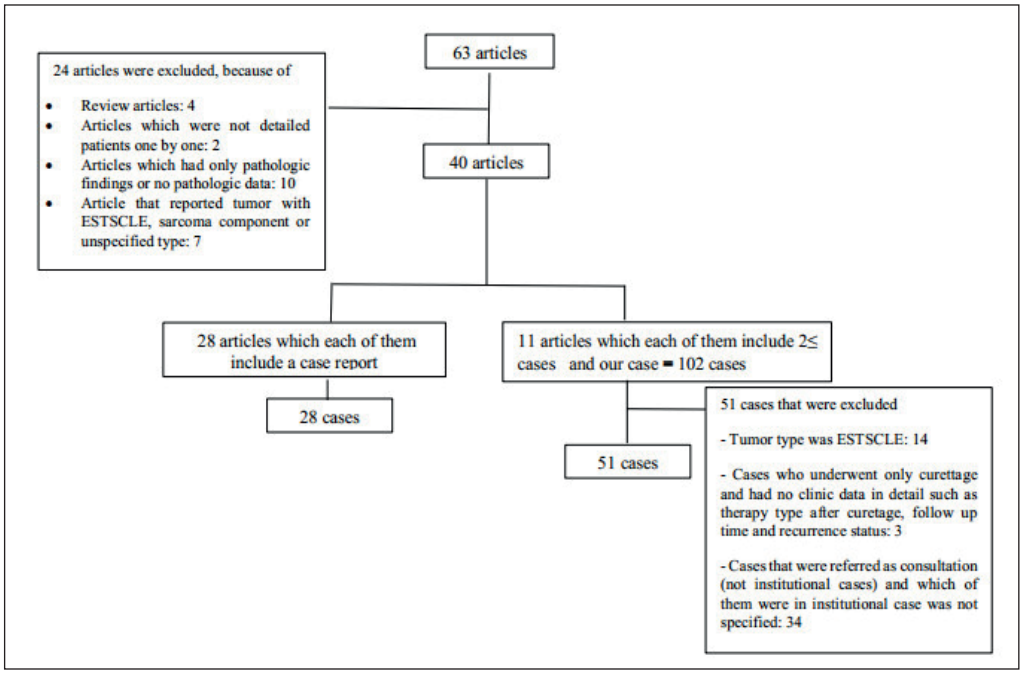

Figure 6: Chart of the literature electronic search of UTROSCT (Abbr. in figure: ESTSCLE; endometrial stromal tumors with sex cord-like elements).

preserving (all others). Disease-free survival (DFS) was defined as the time period from initial therapy to recurrence or last contact. Time from initial therapy to death or the last contact was defined as median follow-up time.

SPSS 17.0 (SPSS Inc., Chicago, IL) was used for the data management and statistical analysis. Descriptive statistics were expressed as *mean±standard deviation (SD)* or *median (min-max)* for continuous variables and number/percentage for categorical variables. The Kaplan-Meier method was used for the assessment of survival outcomes. Survival curves were compared using the log-rank test. *P*-values less than 0.05 were considered significant.

Results of Systematic Review Analysis

The median age of entire cohort at initial diagnosis was 49 years (range: 16-86 years). Age was under 40 years in 21 (26.6%) patients and ≤30 years in 11 (14%) patients. Four (5.1%) patients were in the adolescent and young adult age range (≤25 years). The most common symptom was abnormal bleeding (67.1%). Other reported symptoms were pelvic pain in 4 (5.1%) patients, palpable pelvic masses in 2 (2.5%) patients and abnormal bleeding with galactorrhoea in 1 (1.3%) patient. Fourteen (17.7%) patients were asymptomatic. Symptoms were unreported in five patients.

One patient received primary radiotherapy. One patient underwent neoadjuvant radiotherapy followed by surgery (hysterectomy and lymphadenectomy). The remaining patients (n: 77) underwent surgery. Surgery was performed as a hysterectomy with or without BSO in 61 patients, hysterectomy with BSO and lymphadenectomy in 7 patients, myomectomy in 2 patients, hysteroscopic mass resection in 6 patients, and transvaginal mass extirpation in 1 patient. Three patients received adjuvant therapy. One patient underwent adjuvant radiotherapy (external beam radiotherapy and vaginal vault brachytherapy) following hysterectomy and BSO. One patient who underwent surgery (hysterectomy with BSO and lymphadenectomy) received adjuvant chemotherapy (BEP protocol (four cycles): bleomycin, etoposide, and cisplatin) followed by radiotherapy because of the positive surgical margin in the parametrium. One patient who had an iliac lymph node metastasis in the final pathology received adjuvant progestin therapy following surgery (hysterectomy, BSO, and lymphadenectomy).

The median tumor diameter was 50 mm (range: 4-140 mm). In 75 (94.9%) patients, there was a single lesion. There was necrosis in 4 (5.1%) patients and atypia in 16 (20.3%) patients. At least one mitosis per 10 HPF was determined in 36 (45.6%) patients. In three cases, the mitotic index was 8, 9, and 11 per 10 HPF. In four cases, the mitotic index reported as <10 per 10 HPF. In the remaining cases with mitosis, it was <5 per 10 HPF. The tumor border was infiltrative in 34 (43%) patients. A pushing pattern with a well-circumscribed border was determined in 28 (35.4%) patients. The tumor was confined to the endometrium in 18 (22.8%) patients, involving at least part of the myometrium but no serosa in 46 (58.2%) patients, and involving all layers (serosa included) in 8 (10.1%) of patients. There was cervical involvement in 3 (3.8%) patients. The extra-uterine spread was determined in 5 (6.3%) patients. The locations of extra-uterine spread were in the ovary and appendices epiploicae in one patient, ovary and parametrium in one patient, appendices epiploicae alone in one patient, and internal iliac lymph node metastases in two patients. Clinical-pathologic features of the entire cohort are shown in Table I.

Table I: Clinical-pathologic features of entire cohort

Parameters		**n**	**(%)**
Menopausal status	Premenopausal	32	40.5
	Postmenopausal	29	36.7
	NR	18	22.8
Number of lesion	Single	75	84.9
	Multiple (2≤)	3	3.8
	NR	1	1.3
Involved layers of uterus	Confined to endometrium	18	22.8
	At least part of the myometrium but no serosa	46	58.2
	All layers (serosa incl.)	8	10.1
	NR	7	8.9
Border type of tumor	Pushing or well circumscribed	28	35.4
	Infiltrative	34	43.0
	NR	17	21.5
Presence of atypia	Absent	18	22.8
	Present	16	20.3
	NR	45	57.0
Presence of mitosis	Absent	18	22.8
	Present	36	45.6
	NR	25	31.6
Presence of necrosis	Absent	20	25.3
	Present	4	5.1
	NR	55	69.6
Cervical involvement	Absent	76	96.2
	Present	3	3.8
Presence of extra-uterine spread	Absent	54	68.4
	Present	5	6.3
	NR	20	25.3

NR: Not reported.

Table II: Clinical-pathological factors of recurrent patients with UTROSCT

No	Author/ year	Age	Symptom	Initial therapy	Size of tm (mm)	Localization of tumor	Extra-uterine spread	Mitotic index*	VI	DFI (m)	Symptom in recurrence	Recurrence site	Therapy of rec.	FU- time (m)	Status
1	Biermann et al. 2007 (10)	68	PB	TAH+BSO	45	Uterine	no	no	no	48	Intestinal obstruction	Abdominal (*Gastric serosa and intestinal wall*)	Cytoreductive surgery	48	NR
2	O'meara et al. 2009 (34)	35	AUB + galactorrhoea (Prolactin: Normal)	TAH	99	Uterine	no	mild		36	Galactorrhoea +palpable mass	Abdominal + pelvis + subcutaneous	Cytoreductive surgery and Chemotherapy (BEP protocol)	48	NED
3	Chai-Yan Liu et al. 2015 (30)	50	AUB	Submucous Myomectomy	45	Uterine + cervix	no	NR	no	10	Pelvic mass	Pelvis (*uterine mass*)	Surgery (TAH)	60	NED
4	Endo et al. 2016 (16)	62	NR	TAH	NR	Uterine	no	mild		276	Pelvic mass	Pelvis (*pelvic lymph node +pelvic wall infiltration)*	Hormonal therapy$^{\infty}$ + tumoral arterial embolization + cytoreductive surgery (incomplete resection)	296	Stabile disease
5	Presented case	61	Pelvic mass	TAH+BSO	NA	Uterine	no	2		60	Pelvic mass	Abdomen (*omentum and splenic hilum*) + Pelvis	Cytoreductive surgery + adjuvant hormone therapy$^{¥}$ (1 year)→after 3 months without therapy→seconder pelvic recurrence→ cytoreductive surgery	83	NED

UTROSCT: Uterine tumors resembling ovarian sex-cord tumor, **AUB:** Abnormal uterine bleeding, **PB**: Postmenopausal bleeding, **TAH:** Total Abdominal Hysterectomy, **BSO:** Bilateral Salpingo-oophorectomy, **VI:** Vascular invasion, **m:** Month, **NR:** Not reported, **NA:** Not available, **DFI:** Disease Free Interval, **BEP:** Bleomycin+Etoposid+Cisplatin, **FU-time:** Follow up time, **NED:** No evidence of disease.

*per 10 high power fields. $^{\infty}$ Letrozole (aromatase inhibitor) and medroxyprogesterone acetate during 3 months. $^{¥}$ Megestrol acetate and Letrozole.

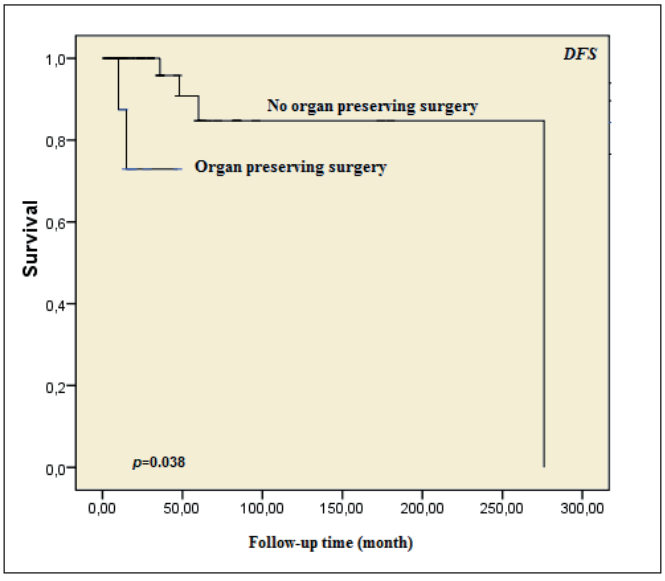

Figure 7: The 5-year disease free survival was 86% and 96% in patients with UTROSCT who underwent organ preserving surgery or not, respectively (p=0.038).

Median follow-up time was 30 months (range: 3-296 months). Recurrence was determined in 5 (6.3%) patients. Median recurrence time was 48 months (range: 10-276 months). The most common recurrent site was in the pelvic region. Recurrences were in pelvis alone for two patients, in the abdomen alone for one patient, in the abdomen and pelvis for one patient, and in abdomen + pelvis + subcutaneous region for one patient. All but one patient with recurrence underwent cytoreductive surgery. One patient (Case 4) received hormonal therapy for three months (letrozole and medroxyprogesterone acetate) and underwent embolization of the tumor arteries followed by surgery. Adjuvant therapy was given as chemotherapy (BEP) to 1 patient (Case 2) and as hormonal therapy (megestrol acetate and letrozole) to one patient (Case 5). Characteristics and pathological findings of patients with recurrent disease are detailed in Table II.

The five-year DFS was 83.4%. The DFS was significantly related only to surgery type. The five-year DFS was

Table III: Association between clinic-surgical-pathologic factors and disease-free survival (DFS)

Parameters		5-year DFS (%)	*p* value
Age	<49	90	0.651
	49≤	81	
Menopausal status	Premenopausal	86	0.810
	Postmenopausal	76	
Tumor diameter (mm)	<50	82	0.441
	50≤	93	
Number of lesions	Single	98	0.841
	Multiple (2≤)	100	
Involved layers of uterus	Confined to endometrium	83	0.792
	At least part of the myometrium but no serosa	96	
	All layers (serosa incl.)	100	
Border type of tumor	Pushing or well circumscribed	89	0.269
	İnfiltrative	100	
Presence of atypia	Absent	100	0.480
	Present	83	
Presence of mitosis	Absent	80	0.653
	Present	66	
Presence of necrosis	Absent	67	0.414
	Present	100	
Cervical involvement	Absent	98	0.649
	Present	100	
Presence of extra-uterine spread	Absent	98	0.360
	Present	100	
Categorization of surgery according to extension	Hysterectomy	87.5	0.969
	Hysterectomy with BSO	82	
	Hysterectomy ±BSO	84	0.681
	Hysterectomy with BSO + Lymphadenectomy	100	
Organ preserving surgery[¥]	Yes	86	0.038*
	No	96	

BSO: Bilateral salpingo-oophorectomy, [¥]organ preserving surgery included patient underwent myomectomy, hysteroscopy, uterine mass excision and transvaginal mass extirpation. *p<0.05 is statistically significance.

86% and 96% for patients who underwent or did not undergo organ-sparing surgery, respectively (p=0.038) (Figure 7). No statistically significant relationship existed between hysterectomy only and hysterectomy with BSO (87.5% *vs.* 82%; respectively, p=0.969) groups. Age *(<49 vs. 49 ≤),* menopausal status, tumor diameter *(*<50 mm *vs.* 50 mm ≤*)*, presence of cervical involvement, presence of extra-uterine spread, border type of the tumor, number of lesions, mitosis, atypia, necrosis and involved layers of the uterus were not associated with DFS (Table III). End status was reported in 56 patients in the literature and no patients died because of the disease.

DISCUSSION

UTROSCT is one of the rarer types of uterine tumors. The differential diagnosis of UTROSCT should consider epithelioid leiomyoma, ESTSCLE, endometrioid carcinoma with sex cord-like elements, carcinosarcoma, and adenosarcoma (44). Even though pathologic features in H&E staining for UTROSCT have been detailed, evaluation with immunohistochemical staining is usually needed to achieve a correct diagnosis. Various immunophenotypes are characteristic of UTROSCT (44). Factors affecting the clinical behavior of UTROSCT remain unknown.

The long-term clinical behaviors of UTROSCT are less aggressive than ESTSCLE (4, 18). The recurrence rate is approximately 13% in sex-cord-like uterine tumors, but most include ESTSCLE (45). A popular belief is that UTROSCT has a clinical pattern like a benign neoplasm (23). However, UTROSCT should be considered as a neoplasm with a low malignant potential because of potential recurrence or extra-uterine spread (4, 6, 44). Although UTROSCT has a low recurrence rate (34), this rate is not clearly specified in the literature. Moore and McCluggage reported eight cases with recurrence and a recurrence rate of 23.5% for UTROSCT (43). Their report is the largest case series documented in the medical literature; however, they attributed the high percentage rate to the inclusion of referred patients with metastases to their institution for consultation. According to our literature analysis, the average recurrence rate was 6.3% for UTROSCT.

The preferable treatment for UTROSCT is surgery (4, 30). However, this treatment remains controversial. Blake et al. reported that the addition of adnexectomy to a hysterectomy was not related to differences in DFS compared to hysterectomy alone (4). Similarly, the addition of BSO or lymphadenectomy in the present study was not associated with improvement in survival. A considerable number of patients with UTROSCT (26.6%) were under 40 years old, including adolescent and young adult patients (5.1%). Because of the high probability of fertility and organ-preserving desire in this age group, a clinical decision about surgery type in UTROSCT can be difficult. Therefore, it is important to determine risk factors for recurrence or extra-uterine disease in this population of UTROSCT. Pradhan et al. reported that infiltrative border, vascular invasion, frequent mitotic figures, serosal rupture, stromal predominance and cytologic atypia were associated with the recurrence of UTROSCT (44). Infiltrative borders, vascular invasion, and mitotic count have been asserted as possible predictive factors for aggressive UTROSCT (23). Moore and McCluggage determined that necrosis and mitotic activity were significantly associated with the malignant behavior of UTROSCT (43). According to our results, statistically significant differences in DFS were only determined between patients with and without organ-preserving surgery. However, no clinical-pathological factors such as age, menopausal status, atypia, necrosis, mitosis, type of border, number or location of the lesion, the diameter of the tumor, or cervical involvement were related to recurrence in our analysis. Although DFS was lower in patients who underwent organ-preserving surgery (5-year DFS 85% vs. 96%), no patients died because of the disease. Complete resection with hysterectomy should be a primary option in UTROSCT. In patients who desire fertility, organ-preserving surgery can be considered after giving comprehensive information about recurrence rates and lower DFS. Patients who choose the conservative management must be followed up closely. Complete surgery must be recommended following childbirth. However, the four patients included in our case experienced disease recurrence, although they had hysterectomies. The role of adjuvant therapy is equivocal and reaching a conclusion is not possible on this issue due to the lack of cases in the literature. Due to the absence of distinct prognostic factors, close follow-up is necessary for all patients.

The main limitations of this study are the small study group size and the retrospective design. Nevertheless, we contributed one case with recurrent UTROSCT to the medical literature. According to our knowledge, our study is one of the largest reports of this phenomenon that has comprehensively evaluated the recurrence pattern and assessed the DFS by considering types of treatment.

In conclusion, the accurate pathologic diagnosis of UTROSCT has an important value in shaping surgical management and management during the follow-up period. Patients with UTROSCT require close follow-up to be aware of recurrence. Surgery type is the only factor as-

sociated with DFS. The main treatment is complete surgery (hysterectomy). However, organ-sparing surgery can be useful for young adult patients and those individuals desiring fertility. Given the poor DFS, patients managed with conservative therapy should be followed up carefully and closely for recurrence.

CONFLICT of INTEREST

The authors declare no competing interest. This research did not receive any specific grant from funding agencies in the public, commercial, or not for profit sectors.

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