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Clinicopathological features of two cases of uterine tumors resembling ovarian sex-cord tumors (UTROSCTs) and a comprehensive review of literature

M. Viau[1], K. Grondin[2], J. Grégoire[3], M.C. Renaud[3], M. Plante[3], A. Sebastianelli[3]

[1] Department of Obstetrics and Gynecology, [2] Pathology Department, [3] Gynecologic Oncology Division, Centre Hospitalier Universitaire de Québec, L'Hôtel-Dieu de Québec, Laval University, Québec City, Québec (Canada)

Summary

Purpose: UTROSCTs are rare neoplasms of the uterine corpus. The authors report the clinicopathological characteristics of two cases of UTROSCTs and review the literature on these tumors. *Materials and Methods:* Medical records of two patients treated for an UTROSCT were analysed. A comprehensive review of literature was also performed. *Results: Case 1:* A 49-year old patient underwent an abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH-BSO) for a suspected leiomyoma. The uterus contained leiomyomas and a 1.8-cm nodule diagnosed as an UTROSCT. Immunohistochemical analysis showed positivity for sex-cord and smooth muscle markers. *Case 2:* A 43-year-old patient underwent TAH-BSO for a uterine tumor adherent to the sigmoid serosa and pelvic peritoneum. Debulking surgery allowed complete cytoreduction. The pathologic analysis was compatible with UTROSCT. Cells stained for sex-cord and epithelial markers. Patient received adjuvant chemotherapy with bleomycin, etoposide, and cisplatin. Forty months after surgery, a three-cm UTROSCT lesion in the posterior cul-de-sac was surgically removed. *Review of Literature:* A review of the literature identified 85 cases of UTROSCTs. Hysterectomy is the gold standard therapy. More conservative surgery is an option for selected patients of childbearing age who wish to maintain their fertility potential. Recurrence or metastases are mainly addressed surgically. *Conclusion:* Due to the scarcity of published cases, there is little definite knowledge about UTROSCTs. UTROSCTs are of low malignant potential, thus long term follow-up is recommended.

Key words: UTROSCT; Uterine tumors resembling ovarian sex-cord tumor; Uterine tumors; Sex cord-like elements; Chemotherapy.

Introduction

Uterine tumors resembling ovarian sex-cord tumors (UTROSCTs) are rare uterine neoplasms [1]. They were first described by Clement and Scully in 1976 [2]. These tumors are part of endometrial stromal and related tumors of the uterine corpus in the 2014 WHO Classification of Tumors of Female Reproductive Organs [1]. UTROSCTs contain sex-cord like elements without recognizable endometrial stroma [1]. The terminology UTROSCT is reserved for tumors essentially or exclusively composed of sex-cord-like areas [3].

Multiple oncogenesis hypotheses have been proposed. These polyphenotypic tumors could possibly derive from pluripotential mesenchymal uterine cells [4-9], endometrial stroma [10, 11], adenomyosis or misplaced ovarian tissue [2, 12]. However, their true origin remains uncertain [10, 13]. They often show a variety of sex-cord, epithelial, and smooth muscle differentiation, and they express different combinations of immunohistochemical markers [5, 13-18].

The authors report two cases of UTROSCTs. Of particular interest, one of the patients experienced a pelvic recurrence 40 months following initial surgery and adjuvant chemotherapy. She subsequently underwent salvage surgery and remains free of disease. A comprehensive review of the literature was conducted to assess the clinical features, histopathological, and immunohistochemical features, as well as appropriate surgical approach and adjuvant treatments relevant to these tumors.

Materials and Methods

Two patients were treated for an UTROSCT at a tertiary care hospital. Medical records from these two patients were thoroughly analysed and written consent was obtained from them.

To perform the review of literature, a search was conducted through PubMed in September 2015 using the keywords ''uterine tumors resembling ovarian sex-cord tumors''. There were no criteria regarding the type of articles, year of publication or language. Clinicopathological characteristics, treatments and outcomes were collected.

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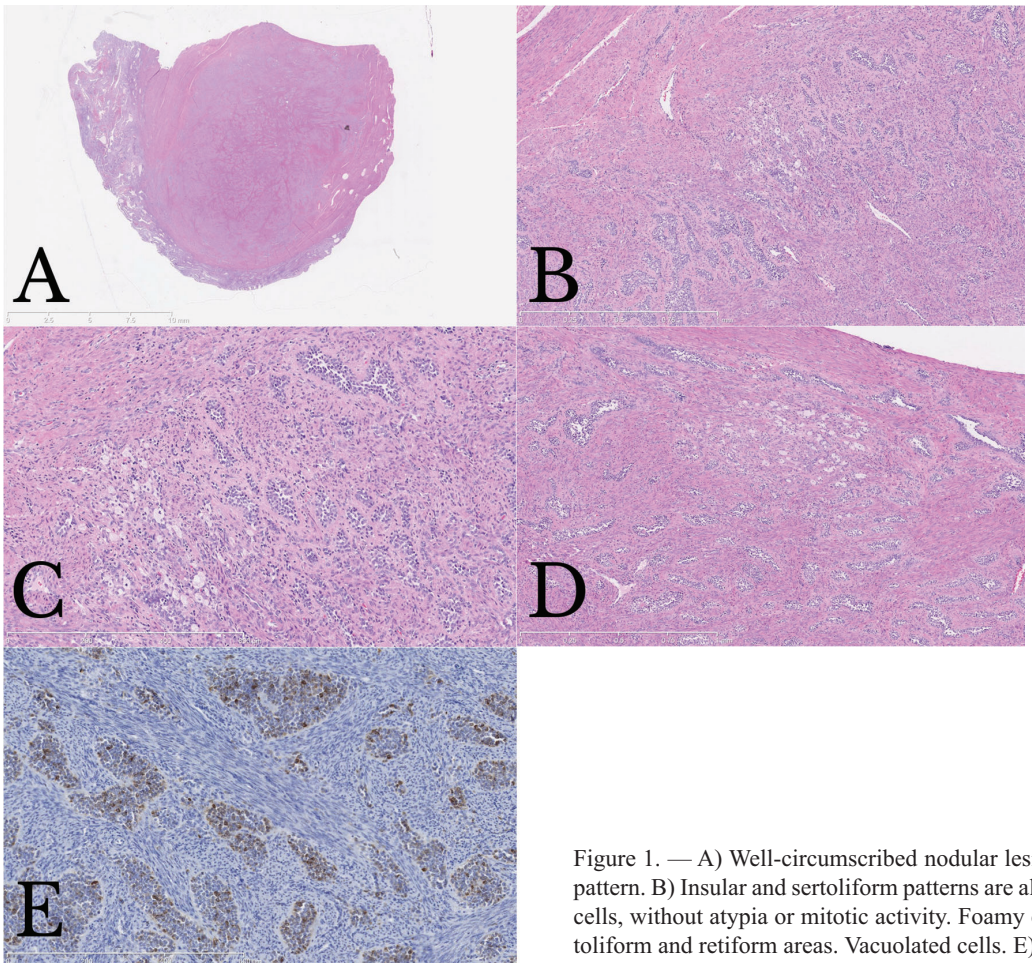

Figure 1. — A) Well-circumscribed nodular lesion with trabecular growth pattern. B) Insular and sertoliform patterns are also present. C) Small round cells, without atypia or mitotic activity. Foamy cells are also seen. D) Sertoliform and retiform areas. Vacuolated cells. E) Inhibin staining.

Table 1. — *Immunohistochemical analysis.*

Marker		Case 1	Case 2
Sex-cord	α-inhibin	+	Focal
	CD99	+	+
	CD56		+
	Calretinin	Focal	+
	Melan-A	Focal	Focal
	Wilms Tumor 1	Weak	+
Myoid	Smooth Muscle Actin	Focal	-
	Desmine	-	-
	h-caldesmone	-	-
Epithelial	Epithelial Membrane Antigen	-	+
	Cytokeratin AE1/AE3	+	+
	Cytokeratin 5.2		+
Miscellaneous	CD10	-	+
	Vimentin		+
	Human Melanoma Black 45		-
	S-100		-
	CD117		-
	Cytokeratin 5/6		-
	Renal Cell Carcinoma		-
	Estrogen receptor	+	
	Progesterone receptor	+	

Results

Case 1

A 49-year old patient (gravida 1, para 1) presented with abdominal pain and pollakiuria. The pelvic ultrasound revealed normal ovaries and an eight-cm mass suggestive of uterine leiomyoma. She underwent abdominal hysterectomy with bilateral salpingo-oophorectomy. Immediate postoperative evolution was unremarkable. She remains radiologically disease-free at 16 months post-surgery.

On gross inspection, the uterus contained numerous lesions with typical appearance of leiomyomas. Apart from these, a yellow 1.8-cm well circumscribed nodule was identified in the myometrium. Microscopic examination of this nodule showed a trabecular growth pattern with sertoliform and retiform areas (Figure 1). Foam cells were present. There was no atypia, mitosis, necrosis or lymphovascular infiltration. Ovaries were normal. Immunohistochemical analysis showed positivity for sex-cord markers, smooth muscle markers, and estrogen and progesterone receptors (Table 1).

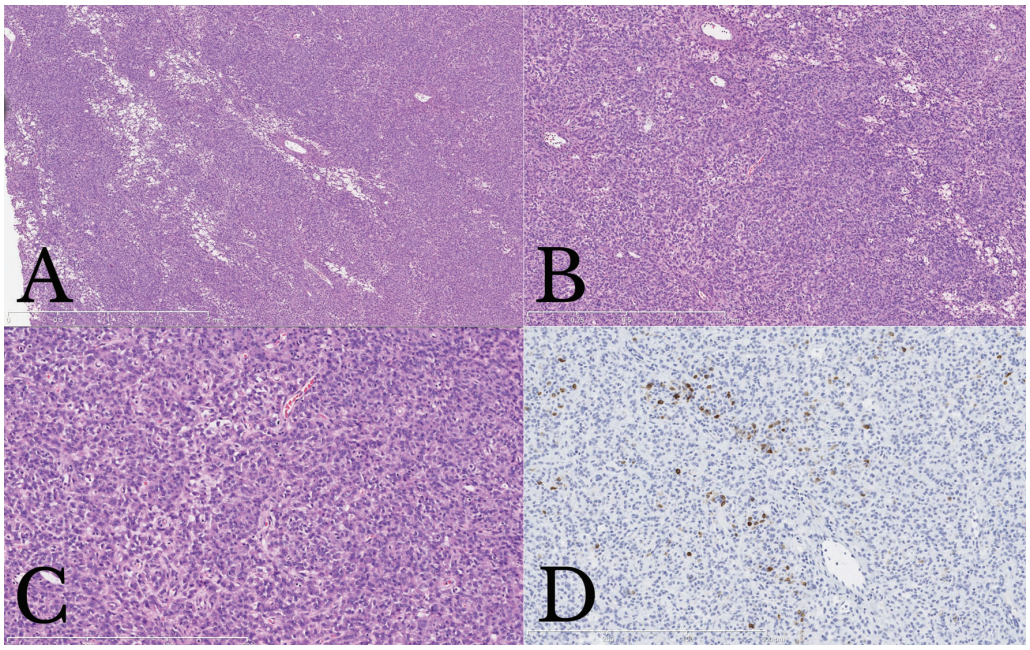

Figure 2. — A) Solid architecture. B) Slightly trabecular growth pattern with cells containing foamy cytoplasm. C) Hyalinized stroma. Small round cells without atypia or mitotic activity. D) Focal positivity for inhibin.

Case 2

This patient first presented in 2010 at age 43 with a three-month history of persistent pelvic pain and pollakiuria. Pelvic ultrasound demonstrated a 15-cm complex paramedian pelvic mass, contiguous to the left side of the uterus. A pelvic MRI described the mass as complex with both fluid-filled loculations and enhancing solid portions. There was also a small calcified nodule at the uterine fundus, consistent with leiomyoma. No pelvic adenopathies were detected, and there was modest amount of pelvic ascites.

Preoperative CA-125 level was elevated (186 kU/L), while CEA was in the expected range for a smoker. This patient underwent an exploratory laparotomy with peritoneal washings followed by an abdominal hysterectomy with bilateral salpingo-oophorectomy through a median incision. A pedunculated solid mass was found to be attached to the uterine fundus. The mass ruptured during the procedure. There were macroscopic implants on the sigmoid serosa and on the pelvic peritoneum which were surgically removed. Omentectomy and pelvic lymphadenectomy were also performed. Surgery allowed complete tumor debulking.

Macroscopically, the pedunculated mass was homogenous, firm, and measured 13 cm in maximum width. Its surface was smooth, but ruptured. Its color ranged from whitish to yellowish. A second tumor, mostly located in the uterine wall (subserosal), was found in the anterior myometrium. This yellowish solid mass measured 5.5 cm and had irregular margins. Both tumors had the same morphologic appearance. Architecture was mostly solid, with trabecular area and a highly hyalinized stroma (Figure 2). Foam cells were seen. The tumor cells were positive for sex-cord and epithelial markers, but negative for smooth muscle markers (Table 1).

Both tumors were diagnosed as UTROSCTs. Pathology also confirmed peritoneal and sigmoid involvement. Ovaries were normal. The rest of the staging was otherwise negative. Diagnosis of a UTROSCT Stage IIB based on FIGO 2009 uterine sarcoma staging system was established. Extrapolating from the data available in the literature on the treatment for sex-cord tumors of the ovary, the present authors' multidisciplinary tumor board suggested adjuvant chemotherapy and the patient underwent three cycles of BEP (bleomycin, etoposide, cisplatin). Following surgery and chemotherapy, serum CA-125 level normalized and radiologic follow-up was unremarkable.

Forty months after initial surgery, the patient complained of leg pain, early satiety, and rectal pain with a change in her bowel habits. A CT scan revealed a three-cm enhancing right mass in the posterior cul-de-sac without a cleavage plane with the rectosigmoid. There were no suspicious lymph nodes. A PET scan showed high metabolic activity in the pelvic mass and no evidence of distant metastasis. Preoperative tumor markers CA-125 and CA 19-9 were normal, and CEA was still within the expected range for a smoker.

Diagnostic laparoscopy showed an isolated, central pelvic mass in the posterior cul-de-sac with no evidence of upper abdominal involvement or carcinomatosis. A rectosigmoid low anterior resection through a median laparotomy allowed complete tumour excision.

The tumor's maximal width was 5.5 cm. The macroscopic appearance was the same as the UTROSCTs diagnosed on the hysterectomy specimen, with no evidence of

significant cytologic atypia or mitotic activity. The case was submitted for a second opinion to an outside reference pathologist who confirmed the diagnosis of UTROSCT.

Peritoneal washings and lymph nodes were negative. The patient received no further treatment and was offered close follow-up. Six months following surgery, all tumor markers remained within normal levels. The patient is actually disease-free two years after salvage surgery.

Discussion

UTROSCTs are rare neoplasms. Previously, the term UTROSCT was also used to describe endometrial stromal tumors with sex-cord-like elements [2]. It is however now restricted to tumors with pure or prominent patterns seen in sex-cord-stromal tumors of the ovary [1]. A review of the literature identified 85 cases fulfilling the current definition.

The most common related clinical presentations are postmenopausal bleeding, abnormal vaginal bleeding, and pelvic pain [2, 4], although some are found incidentally [1, 3]. Rare paraneoplastic syndromes were also reported [19, 20]. UTROSCTs usually arise during the perimenopausal period [3, 15, 21] with a median age at presentation of 50 [1, 4]. There are no well-defined tumor markers associated with UTROSCTs, although elevated CA-125 level has occasionally been reported [19, 22, 23].

Macroscopically, UTROSCTs generally present as an intramural, subserosal, or submucosal uterine mass [1-3, 15, 24]. Two cases [20, 25] of cervical UTROSCTs have been reported. The mean diameter is six cm [1, 3, 21]. On cut section, they usually present as a yellow to tan colored soft and fleshy well-circumscribed mass [3, 5, 6, 15, 21].

The usual histologic features show pushing or infiltrating borders [1-3, 13, 21, 24]. Mitotic activity is generally low [1-4, 13, 15] and nuclear atypia is rare [1, 14]. Necrosis [2, 15] and lymphovascular space invasion are usually absent [1, 4, 13, 14].

There is a variable expression of epithelial, smooth muscle, and sex-cord markers as a consequence of these tumors polymorphism [8, 13, 15, 17]. They are generally positive for at least one sex-cord-stromal marker such as calretinin, inhibin, CD99, Melan-A, WT1 or CD56 [4, 5, 15]. These markers are variably expressed. Their expression in UTROSCTs is consistent with a true sex-cord differentiation [6, 7, 26]. CD99 [27] and CD56 seem to be the most frequent markers of sex-cord differentiation in UTROSCTs. Epithelial components frequently show positivity for cytokeratins [1, 4, 5, 13, 15, 21], while EMA is usually negative [13, 24, 26]. Desmin and smooth muscle actin (SMA) are frequently expressed [1, 21]. h-caldesmon is usually negative [5, 13, 17, 24, 26]. CD10 shows low expression rates [4, 21]. Table 2 lists the available immunohistochemical studies, with the proportion of tumors positive for each marker. These findings are consistent with previous analyses [4, 5].

Definite identification of these tumors can be challenging, thus the use of immunohistochemical panel is essential [6, 17]. Differential diagnosis of UTROSCTs includes endometrial stromal tumors with sex-cord elements (ESTSCLEs), endometrioid carcinomas with sex-cord-like features, epithelioid smooth muscle tumors, PEComa [6, 24], plexiform tumorlet [2], Müllerian adenosarcoma with sex-cord-like differentiation [28, 29], and metastatic ovarian granulosa-cell or Sertoli-cell tumor [5, 30]. However, these tumors show histological characteristics and immunohistochemical markers distinct from UTROSCT that allow correct diagnosis.

UTROSCTs are generally of low malignant potential [2, 4-6, 26, 31-34]. However, considering the small number of cases and limited available follow-up data, the long-term clinical behavior of UTROSCTs remains to be established [2, 4, 6, 14, 26, 35]. The diagnosis of UTROSCT calls for close follow-up [2, 15, 19]. Prognostic factors associated with recurrence include uterine serosal infiltration [19] or rupture, vessel invasion, stromal predominance, and cytologic atypia [21].

Hysterectomy is the gold standard initial therapy for tumors confined to the uterus. It is unclear if bilateral oophorectomy is beneficial. Only two cases with lymph node involvement [36, 37] have been described. It is therefore unknown whether lymphadenectomy is required in the staging procedure and thus the decision is at the surgeon's discretion.

Although uterine mass removal alone with clear margins can be offered in the absence of poor prognostic factors in younger patients wishing to preserve child-bearing potential [2, 4, 14, 38], these patients should be informed of a potential risk of locoregional recurrence. Hysteroscopic resection has been achieved in few cases [14, 32, 34, 39]. Two pregnancies have been reported after fertility sparing surgeries, respectively endoscopic polyp resection [32], and laparoscopic myomectomy with negative margins [38]. Definitive surgery with hysterectomy should be discussed at the end of patient's child-bearing period, as late relapse is possible.

In some reported cases, adjuvant treatments were added to surgery. As most of these tumors have progesterone receptors [4, 9], high-dose progestins have been used after surgery [31, 37] and at the time of recurrence [40]. Use of pelvic radiotherapy with brachytherapy is anecdotal, as it has only been reported once in the neoadjuvant setting [25], and twice as adjuvant therapy [40, 41]. The paucity of cases precludes drawing of any conclusion regarding the effectiveness of these treatments.

Only a few cases of metastatic UTROSCTs have been published. They all have been addressed surgically; two cases [37, 42] with omental metastases, and one [36] with

Table 2. — *Summary table of common immunohistochemical stainings for UTROSCT available in the literature.*

	Sex-cord						Myoid			Epithelial				Miscellaneous				
Reference	Inhibin	Calretinin	CD99	CD56	Melan-A	WT1	SMA	Desmin	h-caldesmon	Cytoketarins AE1/AE3	CAM 5.2	EMA	Vimentin	CD10	HMB45	S-100	ER	PR
Abdullazade [5]	2/3	2/2		2/2				2/2	0/2	2/3		0/3		0/2				
Abid [44]	1/1	1/1	1/1					1/1		1/1			1/1	0/1				
Anastasakis [32]	1/1	1/1					1/1	1/1			1/1		1/1	0/1			1/1	1/1
Baker [26]	5/5		5/5				2/5	1/5		4/5	4/5	0/5						
Bakula-Zalewska [31]	2/4	4/4					2/4	1/4	0/4	2/4				4/4				4/4
Berretta [14]	1/1	1/1	1/1		0/1	1/1	1/1	1/1	0/1	1/1				0/1				1/1
Biermann [33]	1/1	1/1	1/1	1/1	0/1		0/1			1/1		0/1	1/1	0/1	0/1		1/1	1/1
Czernibilsky [7]	1/1	1/1	0/1		0/1			0/1	0/1	1/1		0/1	1/1	0/1			0/1	1/1
de Leval [13]	2/12	4/12			1/11	4/12	11/12	10/12	1/12			1/11		6/12	0/11			
Franco [45]	0/1						1/1	1/1		1/1	1/1		1/1				1/1	1/1
Garuti [39]	1/1	1/1	1/1				0/1	0/1		1/1		0/1			0/1		1/1	
Giordano [34]	2/2	2/2	1/1					1/1		2/2				1/2				
Giutierrez [46]	1/1	1/1	1/1		1/1		1/1	1/1		1/1		1/1	1/1	0/1	0/1		1/1	1/1
Hashmi [30]	0/1	0/1	1/1		0/1		0/1	1/1		1/1		0/1	1/1	0/1	0/1	1/1		
Hauptmann [47]	0/1		1/1		0/1		1/1	0/1		1/1		1/1	1/1			0/1	0/1	1/1
Hillard [38]	1/1	1/1						1/1		1/1		0/1		1/1				
Horn [48]								1/1		1/1		0/1	1/1			0/1		
Hurrell [8]	2/4	4/4	2/4	4/4	1/4	4/4	4/4	3/4	1/4	3/4		4/4	4/4	0/4	0/4		4/4	4/4
Irving [41]	2/5	5/5	4/5		2/5			3/5			4/5		5/5	4/5			3/5	4/5
Kabbani [25]	0/1	1/1					1/1	1/1		1/1		0/1		0/1				
Krishnamurthy [9]	3/7		7/7		4/7		1/7	1/7		4/7	2/7		6/7				7/7	7/7
Liu [43]	3/4	2/4	4/4			1/4	2/4	2/4		4/4				2/4			3/4	3/4
Macak [36]	0/1	0/1	0/1			1/1	0/1	0/1		0/1				0/1	0/1		0/1	0/1
Motiwala [49]	0/1		1/1				0/1	0/1		1/1		0/1	1/1	1/1			0/6	5/6
Nogales [16]	4/6	4/6		5/6			0/6	0/6			5/6	1/6	5/6	0/6				
Oliva [17]	1/7		4/7					5/7	0/7	5/7				5/7				
O'Meara [19]	1/1	1/1	1/1	1/1			1/1	1/1					1/1				1/1	1/1
Sitic [23]	0/1		1/1		0/1		1/1	0/1		0/1		0/1	1/1	1/1		0/1		
Stefanovic [50]							1/1			1/1			1/1				1/1	1/1
Sutak [51]	0/1	0/1	1/1			1/1	0/1	0/1		0/1	0/1			1/1		0/1		0/1
Suzuki [20]	0/1		1/1				0/1	0/1		1/1	1/1		1/1	0/1				
Umeda [37]	1/1	1/2	2/2	2/2	1/2	2/2	0/2	0/2	1/2	2/2		0/2	2/2	1/2	0/2	1/2	2/2	2/2
Viau Case 1	+	+	+		+	+	+	-	-	+		-		-			+	+
Viau Case 2	+	+	+	+	+	+	-	-	-	+	+	+	+	+	-	-		
Total	40/80 (50%)	40/56 (71%)	43/51 (84%)	16/17 (94%)	12/38 (32%)	16/27 (59%)	32/62 (52%)	39/79 (49%)	3/35 (9%)	45/58 (78%)	19/28 (68%)	9/44 (20%)	37/39 (95%)	28/64 (44%)	0/23 (0%)	2/8 (25%)	27/39 (69%)	39/44 (89%)

SMA: smooth muscle actin; EMA: Epithelial membrane antigen; CAM 5.2: cytokeratin 5.2; HMB45: Human melanoma black;
WT1: Wilms tumor; ER: estrogen receptor; PR: progesterone receptor.

lymph node metastasis. Another case with a positive pelvic lymph node at initial surgery subsequently received high-dose progestin therapy [37]. All these patients have remained disease-free, ten months to eight years after treatment.

Rare cases of locoregional recurrences have been reported [19, 33, 40, 43]. They occurred up to six years after initial surgery. The cornerstone of treatment in these cases was secondary cytoreduction. One patient [40] was offered adjuvant radiation therapy and subsequent high-dose progestin therapy. Another patient [19] was surgically treated, and then received adjuvant chemotherapy with BEP. In the present series, the late recurrence experienced by the patient in Case 2 was treated by surgical resection only. It most likely occurred secondary to microscopic residual disease on the sigmoid serosa. This emphasizes the importance of complete surgical debulking, as the recurrence occurred despite adjuvant chemotherapy. So far, the patient remains disease-free two years after salvage surgery.

Conclusion

UTROSCTs are rare uterine tumors with evidence of true sex-cord differentiation. They have initially been considered in the spectrum of endometrial stromal tumors with sex-cord-like elements, but some clinical and pathological data suggest that they are different neoplasms. More data is required to improve knowledge on prognostic factors, clinical evolution, and optimal management.

Based on a review of the current literature, the gold standard treatment is total hysterectomy with or without bilateral salpingo-oophorectomy. For younger patients of childbearing age, conservative surgical excision with negative margins might be an option considering the usually indolent course of this disease. Nonetheless, UTROCTs should be considered of low malignant potential and close follow-up is warranted.

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Corresponding Author:
A. SEBASTIANELLI, M.D.
Gynecologic Oncology Division
Centre Hospitalier Universitaire de Québec
L'Hôtel-Dieu de Québec
11 Côte du Palais
Québec City, Québec G1R 2J6 (Canada)
e-mail: alex_sebastianelli@hotmail.com