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Case Report

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Inhibin alpha as a tumor marker for early detection of uterine tumors similar to ovarian sex cord tumors

Abstract

Background: Uterine tumors resembling ovarian sex cord tumors (UTROSCT) are a rare type of endometrial sarcoma characterized by uterine stroma. These tumors mimic reproductive cord structures and typically exhibit benign behavior. A thorough evaluation of histological and immunohistochemical features is crucial for accurate diagnosis and effective management of these cases.

Case presentation: This report discusses a 45-year-old Iranian woman who presented with a large uterine mass and subsequently underwent a total hysterectomy and bilateral oophorectomy. Pathological analysis identified the lesion as a uterine sac. Notably, there was a significant difference in Inhibin A levels before and after surgery, suggesting that the tumor was responsible for the secretion of this factor.

Conclusion: We recommend that during the initial assessment of similar patients, the levels of this factor should be included in routine blood tests.

Keywords: Uterine tumors, malignancy, salpingo oophorectomy, case report

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Key messages

This case study demonstrated the importance of distinguishing more details of UTROSCT to recognize it from potentially malignant types. Also, observing this case with Inhibin A changes before and after tumor dissection encourages us to consider the importance of measuring this potential marker for early detection of similar cases.

Background

UTROSCT are extremely rare tumors of the uterus, and their clinical characteristics are not completely understood. Type I tumors have the potential to be malignant and the prognosis is influenced by the extent and stage of any stromal tumor development.[1] Type II, often referred to as classic URTOSCT, is characterized by low malignancy and is generally less concerning due to infrequent recurrences. However, it typically exhibits favorable biological behavior. To identify this specific tumor type, tests for Calretinin and Inhibin positivity, along with CD99 or Melan A staining, are conducted.[2] While all benign, some type 2 tumors can be large and have infiltrative margins. In contrast, type 1 tumors tend to be more aggressive, larger, and associated with more ectopic metastases and a higher recurrence rate.[2]

Case presentation

The case involved a 45-year-old Iranian woman (gravida 2, para. 2) with no significant family history of cancer. She presented to a physician with lower abdominal pain and was subsequently referred to Kamali Teaching Hospital for further evaluation and treatment. An ultrasound of the right uterus and ovary revealed multiple cysts. Magnetic resonance imaging (MRI) showed a lobulated cystic mass measuring 80mm by 110mm in the left lateral body and fundus, which contained a few solid components measuring up to 46mm by 16mm. This solid component exhibited abnormal restriction on the Diffusion-weighted imaging (DWI) and apparent diffusion coefficient (ADC) sequences, suggesting the possibility of a degenerated myoma (Figure 1A & B). A computed tomography (CT) scan indicated a cystic lesion with thick septa and enhancement, originating from the uterus, measuring 110mm. While serum levels of tumor markers such as cancer antigen 125, carbohydrate antigen 19-9, carcinoembryonic antigen (CEA), and lactate dehydrogenase (LDH) were within normal limits, it was noteworthy that Inhibin A levels were elevated (Table 1). Cytological examination of the cervix and endometrium revealed no tumor lesions. The patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy. During the macroscopic examination, the specimen showed significant uterine involvement, containing a tumor measuring 15 cm × 12 cm from the lateral wall near the basal area, which was not connected to the endometrium. There was also 300 cc of yellow fluid; upon aspiration, the tumor size decreased, and it was reported to be 6 centimeters at its maximum diameter (Figure1C & D). The patient signed the consent for the operation or data publication

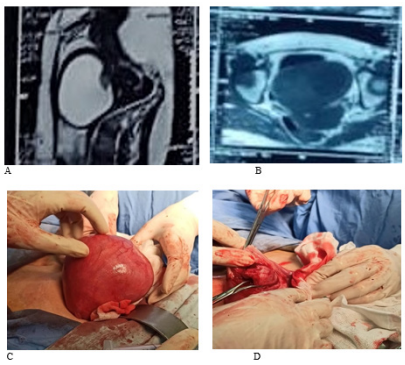

Figure 1 MRI images of the patient before surgery which is a sign of high tumor volume (A, B), Tumor removal (C, D).

Table1 The serum levels of Inhibin A

Test	Result	Unit	Normal range
Inhibin A	399	Pg./ml	Females: Early follicle: 5-28 Mid " : 8-34 Late " : 19-102 Mid cycle : 50-155 Early luteal : 36-133 Mid luteal : 13-160 Late luteal : 7-90 IVF peak levels: 354-1690 Post-ovulatory: 6-16 Postmenopausal:<3.8 Male:<3.5

Inhibin-A concentration in pregnant women:	
Weeks of gestation:	Concentration:
10	101-291
11	89-278
12	87-253
13	62-212
14	74-295
15	73-261
16	54-189
17	59-200
18	67-291
19	61-277
20	74-327

Tissue processing

The standard hematoxylin and eosin staining technique was used to analyze the patient's tissue in the pathology lab. Histological examination showed elliptical spindle cells with varying nuclei and prominent nucleoli within the tumor, along with eosinophilic or sparse cytoplasm exhibiting different patterns. Most of the tumor cells were of mesenchymal origin rather than from the endometrial stroma, and there were no indications of atypia or mitosis. Additionally, a polyp was noted in the cavity of the endometrial lesions (Figure 2). Leiomyomas and serous adenomas were identified as fibroids in the uterus and tumors in the right ovary. No other tumors were detected in the uterus or appendages.

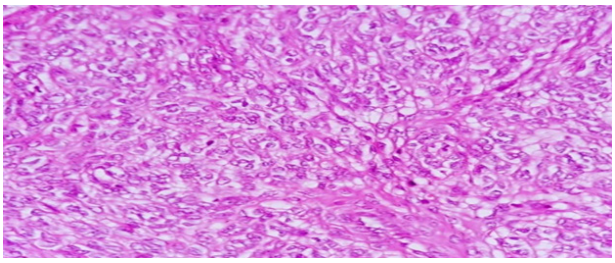

Figure 2 Tumor image after hematoxylin and eosin staining.

Immunohistochemistry

Using immunohistochemistry (IHC), the analysis revealed that the samples tested negative for calretinin, estrogen, and progesterone receptors, as well as for vimentin expression. Neoplastic cells were positive for CD10, Desmin A, SMA (smooth muscle actin), CD56, and WT1 (Wilms tumor protein 1). The Ki-67 labeling index was less than 1%. Additionally, tumor cells were found to be negative for EMA (Epithelial Membrane Antigen), positive for H-caldesmon, and CD99 showed a dispersed positivity in the neoplastic cells.

Discussion

UTROSCT is a rare mesenchymal tumor with an unknown cause.[3] This tumor can be found either intramural, as seen in our case, or in the subserosa of the uterus. Typically, UTROSCT exhibits significant growth, which may or may not involve the invasion of surrounding tissues.[3] It can be challenging to differentiate the tumor from low-grade sarcoma and stromal fibroids when examining it histologically. The diagnosis of UTROSCT relies on a morphological assessment of epithelial cells, aqueous fibers, microcapsules, and dispersed growth patterns.[1] Research indicates that the immunopheno types associated with the umbilical cord provide crucial information. Some sources highlight that, alongside calretinin positivity, a combination of six IHC markers—Calretinin, Inhibin, CD99, CD56, Caldesmon, and WT1—is required for a tumor to be classified as UTROSCT.[3] In this case, similar to ovarian granulosa cells, the tumor marker alpha Inhibin was elevated. Our team monitored the patient for two months following the hysterectomy and observed a decrease in Inhibin A levels after the tumor was removed. Consequently, we propose that alpha Inhibin should be considered a potential tumor marker for diagnosing these types of tumors. Additionally, due to its unique characteristics, UTROSCT can be classified into low-grade endometrial stromal sarcoma with genital cord elements, epithelioid leiomyoma, and endometrioid carcinoma that share similar features with the genital cord, as well as adenosarcoma and carcinosarcoma. These neoplasms exhibit an unusual clinical progression that warrants treatment with less invasive and more considerate approaches. A hysterectomy alongside a salpingo-oophorectomy, as done in the current case, or the excision of a hysteroscopic tumor is a suitable option. Some specialists advocate for the removal of local lymph nodes in the pelvic region or para-aortic lymph nodes. Malignancies associated with UTROSCT are extremely rare, with a low likelihood of ectopic development, metastasis, or recurrence. However, some studies indicate that these tumors can spread to lymph nodes and tissues outside the uterus {Lin,

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2024 #28}. Thus, researching patients with UTROSCT to prevent potential occurrences is particularly crucial. To monitor the condition of UTROSCT patients, it's important to assess Inhibin alpha levels following surgery. An increase in this hormone post-operation may indicate the retention of tumor fragments and potential growth, recurrence, or disease progression for various reasons. Therefore, to establish informative markers for evaluating and understanding a UTROSCT patient's status, we recommend measuring Inhibin alpha alongside Inhibin beta in more cases in future studies.

Acknowledgments

None.

Conflicts of interest

The authors declares that there are no conflicts of interest.

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