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Case Report
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Uterine Tumor Resembling Ovarian Sex-Cord Tumors: Challenging to Diagnose before Surgery

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Abstract

In 1976, Clement and Scully (1) first discussed and proposed the concept of "Uterine Tumors Resembling Ovarian Sex Cord Tumors" (UTROSCTs). But before then, the concept of sex-cord differentiation of endometrial stromal tumors has been reported several times in retrospective analysis. So far, there are a few articles about the image description of UTROSCT in the domestic and overseas. This is a very rare, benign or malignant tumor. Mainly occur in premenopausal and perimenopausal women, with onset around the age of 45. The main symptoms are vaginal irregular bleeding, abnormal menstruation, or lower abdominal pain, and some cases have no symptoms.

Introduction

The clinical manifestations and imaging features of UTROSCT are non-specific, so they are very easy to be misdiagnosed. Currently, pathological biopsy plays a key role in the final diagnosis. Our hospital encountered a case of UTROSCT, which was diagnosed as uterine leiomyoma 3 years ago and was repeatedly considered as uterine leiomyoma in preoperative hysteroscopy, transvaginal ultrasound and pelvic magnetic resonance imaging.

Case Presentation

A 44-year-old female patient, who had been diagnosed with uterine leiomyoma for more than 3 years. Due to irregular menstruation, she was admitted to the department of gynecology of our hospital on June 23rd, 2022. She presented vaginal bleeding with a small amount of yellowish vaginal fluid, no abdominal pain or other discomfort, negative tumor markers, and no abnormal hormone levels.

Transvaginal ultrasound showed that the uterus size was 9.46 cm × 6.96 cm × 6.90 cm, the shape was irregular and the echo was not uniform, thickness of the endometrium was about 1.07 cm. Sonographer detected several slightly hypoechoic masses in uterus and cervix. In pelvic magnetic resonance imaging (Figures 1A-1D), the uterus and cervix increased in volume which had irregular appearance and local bulges. There were multiple circular nodules and masses in the uterine posterior wall and cervix. Compared with myometrium, T1WI presented isointense, T2WI presented uneven slightly hyperintense/hyperintense, DWI presented slightly hyperintense, ADC presented slightly hypointense. And the lesions were significantly heterogeneously dense enhancement on gadolinium-enhanced T1-weighted images, similar to the degree of myometrium enhancement.

The patient underwent hysteroscopic submucosal myomectomy. During the operation, a uterine myomatoid tissue with a size of about 6 cm × 5 cm × 4 cm was observed in the cervix and protruded to the vagina. There was thickening of the endometrium. The cut surfaces of the lesion were gray or gray-red and it grossly seemed to be tough tissue, about 8 cm × 7 cm × 2.5 cm in size. Immunohistochemical results: CD56 (+), CD99 (+), inhibin-α(-). Histology (Figure 1E) and immunohistochemistry suggested a submucosal mesenchymal tumor of the uterus, considering a uterine tumor resembling ovarian sex-cord stromal tumors.

Discussion

In 1976, Clement and Scully [1] proposed UTROSCT, which was divided into two types based on histopathology and biological behavior: Type I is Endometrial Stromal Tumor with Sex-Cord Components (ESTSCLE); Type II, called UTROSCT, consists of tumors totally composed of components resembling ovarian sex-cord tumors, most scholars have accepted that UTROSCT is only used to diagnose type II. In 2014, the World Health Organization incorporated this disease

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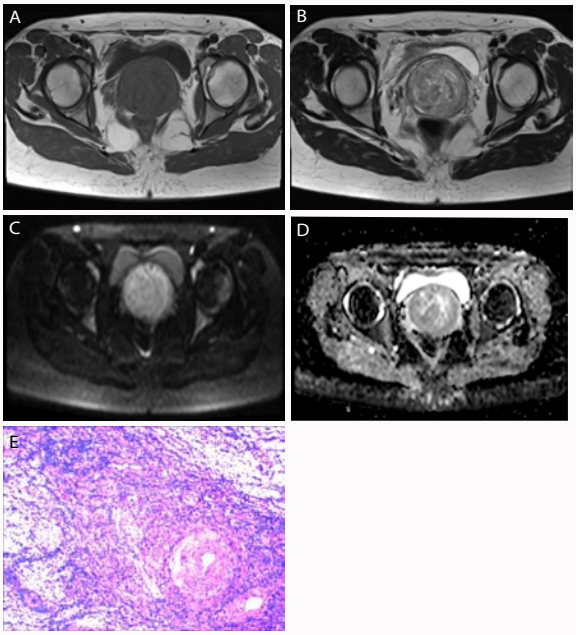

Figure 1: Uterine tumor resembling ovarian sex-cord tumor in a 44-year-old woman.
Pelvic magnetic resonance imaging (A-D) shows multiple circular nodules and masses in the uterine posterior wall and cervix. On T1-weighted image (A), the nodules and masses show iso-signal intensity compared to the myometrium. On T2-weighted image (B), the nodules and masses show uneven slightly hyperintense or hyperintense. There is slight diffusion restriction in the solid portion of the nodules and masses on diffusion-weighted images (C, b = 1000) and the ADC map (D). Microscopic findings (E) of a uterine tumor resembling ovarian sex-cord tumor. The cells were of medium size, with no obvious atypia, scant or empty bright cytoplasm, and dark nuclear staining (hematoxylin & eosin stain, x200).

into the category of endometrial stromal tumors and related tumors, specifically proposing that UTROSCT was a uterine tumor without endometrial stromal components but resembling ovarian sex-cord tumors. In 2020, WHO [2] changed the classification of female reproductive system tumors, defining that UTROSCT is a uterine tumor similar to ovarian sex-cord tumors in shape and there are no endometrial matrix components in this tumor.

The source of UTROSCT is unclear. At present, most scholars believe that UTROSCT contains true sex-cord components, which originates from multipotential mesenchymal cells and those differentiate into sex-cord cells, smooth muscle cells and endometrial stromal cells [3]. Since UTROSCT is characterized by multidirectional differentiation, a variety of uterine tumors with sex-cord differentiation characteristics should be considered in the differential diagnosis of this tumor, such as uterine leiomyoma, endometrial stromal tumor, adenosarcoma, and endometrioid carcinoma and so on. As with endometrial thickening in this case, we need to think of neoplastic lesions associated with hormonal changes, and then take ovarian sex-cord tumors into account. Given that they occur in the uterus, perhaps we should consider UTROSCT. Would the rate of misdiagnosis decrease?

There are few reports on the imaging findings of UTROSCTs. In previous cases, the imaging findings of the tumor were not specific, and most radiologists or physicians did not consider UTROSCT into the differential diagnosis. All UTROSCTs were confirmed by final postoperative biopsy and pathology. In the cases encountered in our hospital, the tumor shows multiple circular nodules and masses located, iso-signal on T1WI and uneven slightly high/high signal on T2WI. Those imaging findings were very similar to uterine leiomyomas. The slightly uneven high/high signal on T2WI is highly consistent with the sign of uterine leiomyoma degeneration. And combined with the multiple characteristics of this case, it is more likely to be misdiagnosed as uterine leiomyoma. In addition, the tumor in this case is characterized by cystic and solid imaging features, which is similar to typical imaging features of granulosa cell tumor. If we associate this subtype of ovarian sex-cord stromal tumor, the diagnostic accuracy may be improved.

Therefore, it is necessary to further study the pathological and imaging characteristics of UTROSCT. At the same time, incorporate UTROSCT into the differential diagnosis of sex-cord differentiated uterine tumors, so as to provide a better basis for clinical diagnosis.

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