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An Ovarian Mass with Microcystic Stromal Tumor: A Rare Case Report

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ABSTRACT

Background & Objective: Ovarian microcystic stromal tumor (MST) is a rare subset of ovarian tumors, that usually appears in the form of a pelvic mass which is often unilateral, and has a microcystic realization characterized by small, elliptical, and circular cysts. This microcystic stromal tumor is a type of ovarian insufficiency that has recently come to researchers' attention. However, no meta-cognitive studies have beem conducted regarding the issue. This tumor morphologically and histologically may look very similar to granulosa cell tumor, Sertoli, Lydic and other ovarian tumors but different characteristic of immunohistochemistry, genetic and gene mutation incidence makes it different. Herein, we report a rare case of the microcystic stromal tumor using immunohistochemistry studies.

Case Report: A 60-year-old woman with ovarian mass referred to gynecology clinic in March 2018. She underwent total abdominal hysterectomy and bilateral Salpingo-oophorectomy. Pathology results showed ovarian microcystic stromal tumor. The patient was fallowed up without any intervention after surgery. To date she is alive with no problem.

Conclusion: Ovarian MST is a rare tumor that originates from the ovarian stroma, which is histologically confused with a number of ovarian tumors, especially metastatic tumors. Immunohistochemistry findings are very helpful in differentiating this tumor from other tumors and preventing diagnostic errors.

Keywords: β-catenin, microcystic, ovarian microcystic stromal tumor

Introduction

Ovarian microcystic stromal tumor (MST) is a rare subtype of ovarian tumors which was introduced by Irving and Yang in 2009. Regarding the incidence of this tumor, the mean age is 45 to 50 years old. These masses usually appear in the form of pelvic masses which are often unilateral. The microcystic realization is characterized by small, elliptical, and circular cysts **(1)**.

The classification of ovarian tumors is mainly based on histological characteristics, which attempt to reflect the embryogenesis and histogenesis of the tumor. According to a pathology outline registered in November 2018, the ovarian MST is defined as a benign ovarian neoplasm. Immunohistochemical findings of the tumor cells indicated positive vimentin and CD10. Furthermore, other epithelial markers, epithelial membrane antigen (EMA), cytokeratin (CK) and sex cord (inhibin and calretinin) were negative. Accordingly, researchers have assumed that ovarian tumors originate from the stroma and emphasized on the unique microcysts as histologic findings of these tumors **(2)**.

Reviewing the literature revealed that only one additional report was carried out by Maeda *et al.* to examine two MSTs. By applying immune-histochemistry, this study indicated the realization of the nuclear unit of β-catenin protein in tumor cells and showed similar point mutations in both cases, demonstrating the role of the dysregulation of the Wnt/β-catenin pathway in the pathogenesis of the MST, showing that β-catenin and Cyclin D1 may also be effective in this pathogenesis **(3,4)**. This article, reports a case of MST using immunohistochemistry studies.

Case Report

This is a case report of a 60-year-old woman (para 3) who referred to Oncology Gynecology department of Mashhad University of Medical Sciences in March 2018 due to abdominal pain. The patient underwent ultrasonography and an ovarian mass was reported with malignant ultrasonographic characteristic and normal tumor markers. According to the mass and abdominal pain, the patient underwent a laparotomy. A sample was taken to carry out the pathology tests and it was diagnosed as MST.

The patient had no traumatic lesions. Chest X ray was normal. According to the ultrasonography, the shape, size, thickness and parenchymal echo of both kidneys were normal. The results of the patient's tests are shown in Table 1.

An adnexal mass with a diameter of 5×5×3 cm was observed. The uterus was normal. In the microscopic findings, parts of the ovarian tissue with neoplastic lesions consisting of small circular microcystic sheets and structures to elliptical spaces accumulated near irregular channels with solid cellular cells. There were no malignancies in the peritoneum. She underwent total abdominal hysterectomy and bilateral Salpingo-oophorectomy. Pathology results showed ovarian microcystic stromal tumor. The patient's histological findings are shown in Figure 1. The patient did not receive any treatment. To date, she is alive with no problem.

Table 1. The tests performed on the patient

TABLE 1 :The tests performed on the patient			
Fasting sugar (mg/dl)	221	Lymphocyte (%)	39.1
Blood urea nitrogen test (mg/dl)	30.4	Alkaline phosphate(IU/L)	236
Creatinine (mg/dl)	1.09	LDL (U/L)	266
Uric Acid (mg/dl)	4.9	CEA(ng/dl)	2.3
White blood cells (ML)	6000	CA-125 (U/mL)	21
Hemoglobin (g/dl)	12.7	SGOT	15
Hemtocrit (%)	38.7	SGPT	14
M.C.V	79	PT patient time	12
M.C.H	25.9	PT control time	12.5
M.C.H.C (g/dl)	32.8	I.N.R	1
Platelet	201000	PTT patient time	40
Neutrophil(%)	54.6		

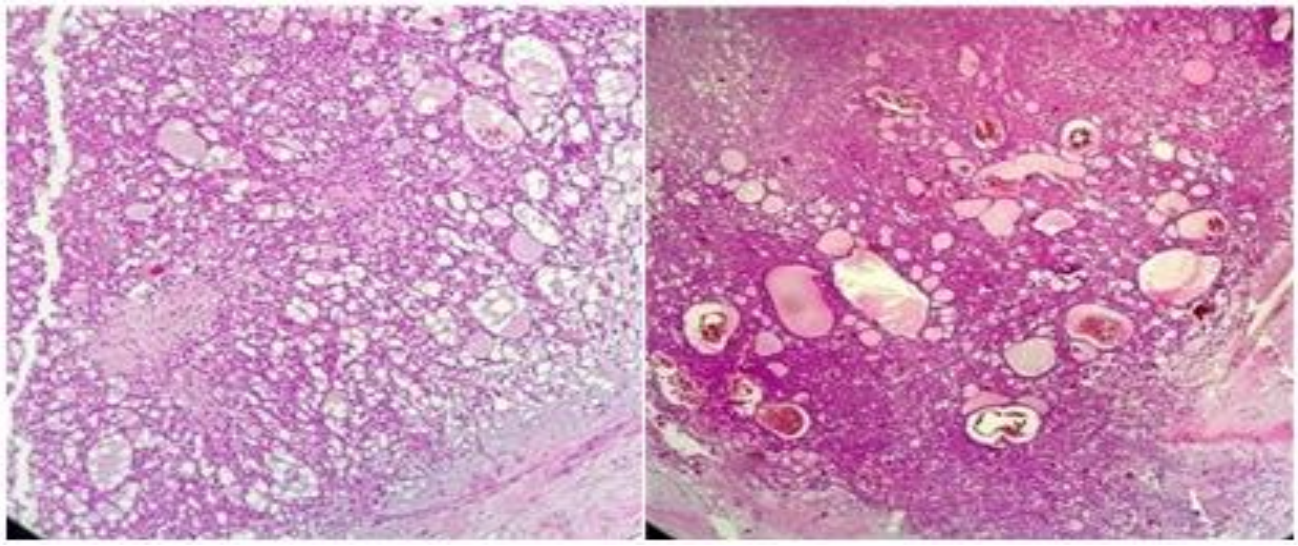

Figure 1. The patient's histological findings: The microcystic structures consisting of non-atypical cubic cells with mitosis and discharges. Sharpness *100. Colored with Hematoxylin and Eosin.

Discussion

Until 2009, few reports of ovarian MST had been presented. Obtaining a specific category distinct from ovarian neoplasms, this tumor was introduced by Irving and Yang in 2009 by reporting a sample of 16 people **(2)**. Based on the histological characteristics, these lesions are apparently reminiscent of thecomas in some areas; however, they have clear microcystic patterns that are not visible in the thecomas. These masses usually appear in the form of pelvic masses which are often unilateral. The microcystic realization is characterized by small, elliptical, and circular cysts. Main characteristics of these tumors include 1) microcystic pattern 2) lack of morphological characteristics in the cervical stromal tumor 3) epithelial elements 4) lack of cellular elements of mass and 5) intracytoplasmic vacuole.

Changes in the ovarian MST are divided into three groups of solid, solid-cystic, and mainly cystic. The case reported by the current study was a cystic mass. Microscopic findings of the ovarian MST usually show the following characteristics: solid, microcystic, or macrocystic patterns, uniform rounded cells with nuclear features, and a few mitosis forms. In addition to the abovementioned microscopic characteristics, a thick hyalinized fibrous band inside the stroma is one of the common microscopic characteristics of MST. Moreover, a thick fibrous band is found in the thecomas **(6)**. The nature of microcysts or macrocysts has not yet been figured out; these cystic characteristics are drawn by tumor cells. However, there is no definitive evidence of lymph nodes or epithelial differentiation **(3,6,13)**.

Unlike epithelial ovarian cancer, sexual steroid hormones of ovarian tumors (including granulosa and thecomas tumor cells) do not have a verifiable hereditary component. Disturbances in regulating the core Wnt/β-catenin pathway may play a role in the development of granulosa cell tumors **(7)**. In addition, imbalances in chromosomes 4, 9, and 12 have been reported repeatedly in thecomas, which indicate that genes in these regions may play a role in the development of these tumors. There is little genetic information for the pathogenesis of sex cord-stromal tumors. The mutation of the DICER1 gene in the Sertoli and Leydig cell tumors **(8)** and the mutation of β-catenin (CTNNB1) S33C have been reported in the ovarian MST **(3)**. According to the previous studies, the origin of MST is still unclear **(2,3)**.

MST has unique histologic and immunohistochemical characteristics. Making a differential diagnosis based on histological findings is essential for a definitive diagnosis. In the differential diagnosis, a wide variety of ovarian tumors, including thecomas, ovarian granulosa cell tumors (AGCTs), stromal sclerosis tumor (SST), Sertoli-Leydig cell tumor (SLCT), yolk sac tumor (YST), and ovarian solid-pseudopapillary neoplasm (SPN), should be considered **(9)**.

Histologically speaking, stromal hyaline plaques and uniform tumor cells are similar findings in thecomas and MST **(6,10)**. However, thecomas are usually followed by a growth pattern of solid cells with no cystic spaces and appear to be yellowish and can be seen at old ages, which is not usually observed in MST. In addition, the estrogen manifestations observed in thecomas have not been reported in MST **(10)**. Furthermore, another difference is the positive expression of inhibin and calretinin in thecomas and their negative expression in MST.

Another case is granulosa ovarian cell tumors which have similar solid and cystic growth patterns. This is while granulosa tumor cells are diagnosed by a short elliptical spindle and internucleus grooves in the ovary. Moreover, spaces filled with eosinophilia fluid derived from granulosa cells are regarded as a unique diagnostic mark for ovarian granulosa cell tumors. Additionally, immunohistochemical staining for inhibin and calretinin is usually positive in ovarian granulosa cell tumors.

Compared with ovarian granulosa cell tumors, which are strongly associated with the mutation of the FOXL2 gene, it has recently been shown that MST is related to the mutation of the β-catenin gene **(11)**. A similar case with the ovarian MST with the exon 3 local mutation of the β-catenin gene was reported by Kang **(9)**. In addition, there are no unique MST microsystems in the stromal sclerosis tumor (SST). Immunohistochemical staining of calretinin and inhibin has always been positive for stromal tumor sclerosis, while it has been negative for MST **(9)**.

Some similar histologic characteristics like microcystic exist in both Sertoli-Leydig tumor and MST. However, they may have typical histological findings relative to Sertoli and Leydig cells. Immunohistochemical staining of calretinin in Sertoli-Leydig cell tumors exhibits positive expression while it is negative in MST. In the case of the yolk sac tumor, a microcystic or reticular growth pattern with primitive tumor cells can usually be seen. The yolk sac tumor has at least a moderate cytological atypia, and increased mitosis and periodic acid-Schiff can be observed in this tumor. The staining of AFP and Glycopan3 is positive for this tumor; however, it cannot be found in MST. In addition, the differential diagnosis of MST from ovarian SPN is important **(12)**. Histologically speaking, MST and SPN have a remarkable similarity in the formation of a solid growth pattern composed of uniform tumor cells. However, pseudopapillary characteristics are not seen in MST. The expression of β-catenin has been reported in both tumors, which means that the single pathway leads to the oncogene of the ovarian tumors **(3,12)**.

Until now, certain immune-histochemistery (IHC) of the ovarian MST have been presented. An analysis of the direct sequence of DNA demonstrated a point mutation of the expression of β-catenin and the iso-histochemical expression of the β-catenin nucleus in

tumor cells. Disruptions of the Wnt/β-catenin signaling process play significant roles in the MST pathogenesis **(9)**. However, molecular studies should be conducted to confirm its contribution to MCT gene tumors.

Conclusion

Ovarian MST is a rare tumor that originates from the ovarian stroma, which is histologically confused with a number of ovarian tumors, especially metastatic tumors. Immunohistochemistry findings are very helpful in differentiating this tumor from other tumors and preventing diagnostic errors.

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Ethical Permission

This study is with patient permission publish her medical data. The identity of the patient was confidential and not disclosed in the study.

Conflict of Interest

The authors declared no conflict of interest regarding the publication of this article.

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