CASE REPORT

Ovarian Mature Cystic Teratoma Combined with Mucinous Borderline Tumor – A Case Report with Literature Review

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ABSTRACT

Mature ovarian cystic teratoma with co-existing mucinous borderline tumor is an extremely rare finding in literature. Here, we report a case of bilateral mature cystic teratoma of the ovary with mucinous borderline tumors and intraepithelial carcinoma in a very young, 26-year-old female. The case was diagnosed histologically through immunohistochemical support. The biological behavior of mature ovarian cystic teratoma with mucinous borderline tumor is between those of benign and malignant tumors, having low malignant potential. Since the patient had given birth to a child, a radical operation was performed and no tumor recurrence was observed after one year follow-up.

KEYWORDS: Ovary, Teratoma, Cystic, Mucinous borderline tumour.

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INTRODUCTION

Mature ovarian cystic teratoma is a benign ovarian germ-cell tumor, which is very common clinically. Characteristically, the mucinous borderline tumor of the ovary belongs to the category of the borderline tumors of the epithelial and stromal surfaces of the ovary. The probability of its occurrence in one ovary is relatively low, and the probability of occurrence of intraepithelial cancer is even lower. Clinical reports on mature ovarian cystic teratoma with mucinous borderline tumors are extremely rare in the literature.[1-3] Here the authors report a case of bilateral mature cystic teratoma of the ovary with mucinous borderline tumors and intraepithelial carcinoma. The findings of this study have provided the insights that will improve the understanding of this severe disease.

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CASE PRESENTATION

This study was approved by the Jinhu County People's Hospital Ethics Committee under Letter No. 20200326. A 26-year-old woman with a history of systemic erythema and received hormonal therapy was admitted to hospital with abdominal pain. Computed tomography (CT) showed huge multiseptated solid-cystic mass with clear boundaries in the bilateral ovaries. The sizes of the masses were 20 cm × 18 cm × 15 cm (left ovary) and 16 cm × 14 cm × 12 cm (right ovary). The serum levels of the carcinoembryonic antigen (CEA) and CA19-9 were 10.06 ng/mL and 61.09 ng/mL, respectively. Intraoperative frozen histological examination diagnosed bilateral mature ovarian teratoma with borderline tumor and intraepithelial carcinoma. Later, radical resection of the tumor was carried out. Generally, the surface of the tumor was smooth, but the cystic mass was partially ruptured, containing a viscous mucus-like substance. The cut surface was multiloculated, with

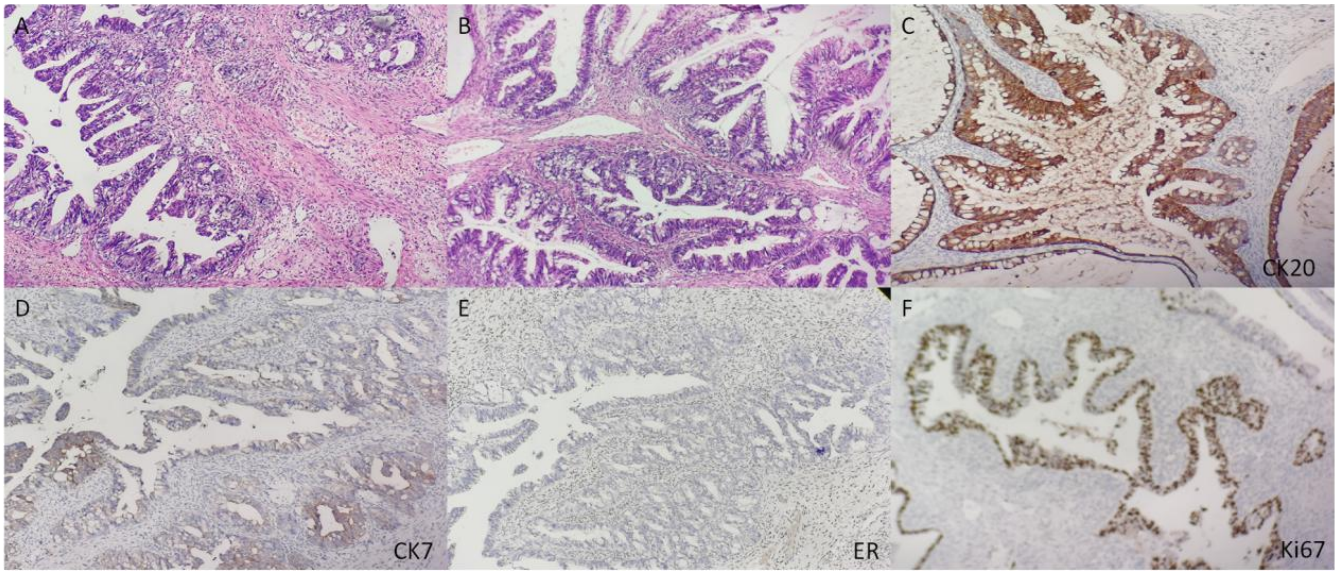

Fig.1: Photomicrographs showing (**A)**Glands are co-walled or form sieve holes; **(B)** The cytoplasm was rich in mucus, and goblet cells were visible; **(C)** These cells were diffusely positive for CK20; **(D)** Cells frequently positive for CK7; and **(E)** Negative for ER; **(F)** The Ki67 proliferation index was approximately 70%.

an intact capsule. The cystic component contained mucus-like substance, and some parts had grease and hair.

Microscopic findings revealed that the mucinous epithelial cells were attached to the tumor sac, and a single layer of benign mucinous cystadenoma changes was present in some areas. The epithelial cells were floating in mucus, infiltrating the ovarian interstitium and forming ovarian pseudomyxoma, with some stratified areas. Interstitial nipples and glands were co-walled or formed sieve holes (Fig.1A). At high magnification, we observed that some of the epithelial cells were well differentiated, with a high columnar shape. The cytoplasm was rich in mucus, with visible goblet cells (Fig.1B). Immunohistochemically, these cells were diffusely positive for CK20 (Fig.1C), frequently positive for CK7 (Fig.1D), but negative for estrogen receptor (ER) (Fig.1E). The Ki67 proliferation index was approximately 70% (Fig.1F).

DISCUSSION

Ovarian mature cystic teratoma with mucinous borderline tumors accounts for only 5% of the cases. In a previous study, the patients were found to be 17–66 years old, with an average age of 39 years.[4] The tumor size varied from 5.5 to 20 cm, and most tumors were unilateral. The case we report here is of a 26-year-old woman with a tumor on both ovaries. Most often, clinical manifestations are abdominal masses with pain.[5] However, symptoms of frequent urination occur in the cases in which the bladder is pressed by the tumor.[6] In this case, a physical examination revealed an abdominal mass, and the patient was admitted to the hospital, usually without symptoms of abdominal pain.

Ovarian mature cystic teratoma with mucinous borderline tumors and intraepithelial carcinoma is often diagnosed after surgery and pathological examination due to its clinical rarity. Mature ovarian cystic teratoma with mucinous borderline tumors and intraepithelial carcinoma usually consists of multi-atrial and smooth masses. Microscopic tumors are composed of cysts and glands of different sizes. The teratoma is can be formed of a single- or a multiple-germ layer. The remaining tumor part is covered with stratified intestinal epithelium, sometimes with papillary protrusions. In our case, the mucous epithelium was benign, and the junction and canceration were visible. Additionally, CK20 was strongly positive, whereas CK7 was focally positive immunohistochemically, which was consistent with the findings of Takao M et al.[7] The vast majority of primary ovarian mucinous tumors originate from

surface epithelial-mesenchymal sources, whereas mature ovarian cystic teratomas derive from primordial germ cells. However, the relationship between the two types is still unclear. Fujii K et al.[8] performed microdissection and genetic analysis of the malignant components associated with mature ovarian cystic teratoma. Their results showed that the malignant components were homozygous genes consistent with those of the mucinous borderline tumor tissue. In another study, Nakatsuka et al.[9] discovered that a premenopausal woman had a mucinous borderline tumor, mature ovarian cystic teratoma, with a transition between gastrointestinal tissue components, which combined the immunophenotypes of keratin and mucin. It currently speculated that the tumor is derived from teratoma gastrointestinal epithelial cells that have undergone malignant transformation. In this case, the patient had been taking hormones for a long time, and thus whether the tumor development is associated with that hormonal therapy remains to be further elucidated.

The biological behavior of mature ovarian cystic teratoma with mucinous borderline tumor is between those of benign and malignant tumors, having low malignant potential. Currently, surgery is the main method of treatment, which depends on the clinical stage at the time of the operation, the age of the patient, and the requirements for fertility.

CONCLUSION

In the present case, we identified a mucinous borderline tumor with mature ovarian cystic teratoma, accompanied by intraepithelial carcinoma, which is particularly rare. Since the patient had given birth to a child, a radical operation was performed. No tumor recurrence has been observed for one year at the time of this manuscript preparation.

LIMITATIONS OF THE STUDY

Due to the few cases diagnosed with this disease, limited treatment experience is currently available. Here, we present only a case report. Although there is no recurrence to date, we still have no experience or standard treatment options to implement.

CONFLICT OF INTEREST

None to declare.

GRANT SUPPORT & FINANCIAL DISCLOSURE

None to disclose.

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Author's Contribution

LW, GC, JJ: Conception and design of study, acquisition of data, drafting of manuscript, revising it critically for important intellectual content.
ALL AUTHORS: Approval of the final version of the manuscript to be published.