



Ovarian endometrioid borderline tumor arising from an endometriotic cyst

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ABSTRACT

Endometrioid borderline tumor of the ovary is defined as a solid or cystic tumor composed of crowded glands lined by atypical endometrioid type cells and lacking destructive stromal invasion and/or confluent glandular growth. In the literature, it was reported to comprise 2-19% of endometrioid tumors and 2-10% of all borderline tumors. The prognosis for ovarian endometrioid borderline tumor is excellent including that with features of intraepithelial carcinoma or microinvasion. Recurrences and metastases are extremely rare. A 34 years old Chinese lady presented with vaginal bleeding for many months. Intraoperatively, an intact right ovarian tumor measuring 55 × 10 × 10 mm was found. Cut sectioned showed a unilocular cyst with hemorrhagic fluid content and a solid tan nodule of 15 mm in diameter was found within the wall. Microscopically, the cystic ovarian tumor showed features of the endometriotic cyst and the solid nodule arising from the cyst wall showed an adenofibromatous pattern of an endometrioid borderline tumor. She also presented with disseminated endometriosis in the omentum and pelvic peritoneum. This kind of case was rarely reported for clinical reference especially on the macroscopic and microscopic features.

KEY WORDS: Endometrioid carcinoma, endometriosis, ovary

INTRODUCTION

Endometrioid borderline tumor of the ovary is defined as a solid or cystic tumor composed of crowded glands lined by atypical endometrioid type cells and lacking destructive stromal invasion and/or confluent glandular growth [1]. Endometrioid borderline tumor is also known as atypical proliferative endometrioid tumor. The microscopic features of this tumor are similar to atypical endometrial hyperplasia [2].

CASE REPORT

A 34 years old Chinese lady presented with vaginal bleeding for many months. Intraoperatively, an intact right ovarian tumor measuring 55 × 10 × 10 mm was found. Disseminated endometriosis was also present. A right salphingo-oophorectomy was then performed. The cut surface of the tumor showed a unilocular cyst with hemorrhagic fluid content and hemorrhagic inner wall. A solid tan nodule of 15 mm in diameter within the cyst wall was seen [Figure 1]. Capsular invasion was absent. Microscopically, the cystic ovarian tumor showed features of the endometriotic cyst and the solid nodule arising from the cyst wall showed an adenofibromatous pattern of an endometrioid borderline tumor. It was composed of crowded endometrioid glands lined by mild atypical columnar cells with grade 1 nuclei within adenofibromatous stroma [Figure 2]. Confluent

glands and infiltrative growth pattern were absent. Squamous metaplasia was seen in abundance [Figure 3]. Disseminated endometriosis was confirmed intraoperatively by the presence of endometriotic foci in the omentum as well as in the pelvic peritoneum.

DISCUSSION

A relatively small number of the ovarian endometrioid borderline tumor had been published but their exact prevalence is uncertain due to variation in diagnostic criteria used. In the literature, it was reported to comprise 2-19% of endometrioid tumors and 2-10% of all borderline tumors [3]. The average age of patients is 51 years where mostly presented with a pelvic mass [1]. This case represents a classical macroscopic and microscopic presentation of an endometrioid borderline tumor of the ovary. The tumor arises from an ovarian endometriotic cyst in the background of multiple endometriotic foci. The association with endometriosis is well established and had been well documented. A study of 38 cases of ovarian endometrioid borderline tumors showed that 14 of the cases were associated with endometriosis [4]. Based on that same study, they also postulated that endometrioid borderline tumor most likely arise from endometriosis whereby the glands become proliferative and atypical resulting in further development into intraepithelial carcinoma, microinvasion and frank invasion [4]. This was proven by a molecular study in

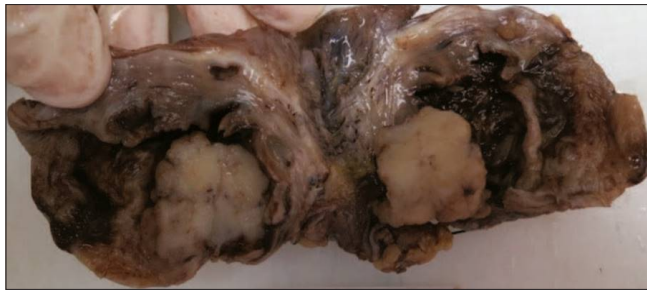


Figure 1: A solid tan nodule attached to the wall of a unilocular cyst

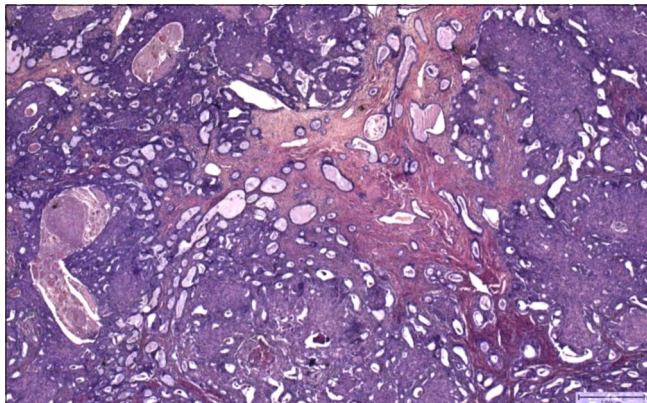


Figure 2: Complex glandular architecture set in a fibromatous background (H and E, x2.5)

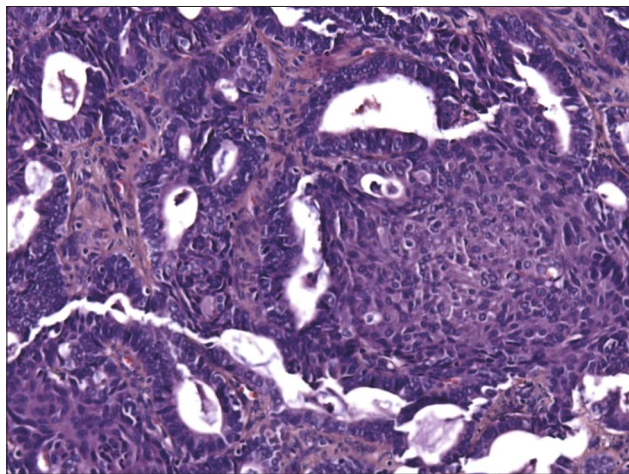


Figure 3: Squamous morules and endometrioid glands exhibiting mild cytological atypia (H and E, x20)

which low-grade endometrioid carcinomas are characterized by mutations in PTEN and CTNNB1, and microsatellite instability, and may arise from ovarian endometriosis or borderline endometrioid tumors [5]. However, high-grade endometrioid carcinomas differ as this entity has similar changes to high-grade serous carcinomas characterized by p53 mutations, and BRCA1 and/or BRCA2 dysfunction [5].

A classical adenofibromatous growth pattern is seen in this case where in about one-half of the cases, an adenofibroma

is present in the background [1]. There are two major histopathological patterns that can be seen in this tumor, which include adenofibromatous and intracystic. A study of 33 atypical proliferative endometrioid (endometrioid borderline) tumors showed that 21 (63%) tumors have the adenofibromatous pattern and the rest showed the papillary (intracystic) pattern [4]. This study also reported squamous differentiation in 16 cases (48%) and the rest showed mucinous differentiation.

As for disseminated endometriosis, in this case, close follow-up is crucial since several studies have reported on the malignant transformation of the endometriotic foci even though the incidence is rare. The ectopic endometrial tissue may undergo malignant change when they are stimulated by factors including intrinsic or extrinsic estrogen [6].

The prognosis for endometrioid borderline ovarian tumors is excellent including that with features of intraepithelial carcinoma or microinvasion. Bell and Kurman (2000) in their clinicopathological study has reported the prognosis of 7 other studies which comprised a total of 106 endometrioid borderline tumors. Out of these cases, 99% showed no evidence of disease within 28-96 months of follow-up [4]. The prognosis is correlated with the findings from the molecular study [5]. Recurrences and metastases are extremely rare. However, for women of childbearing age, unilateral salphingo-oophorectomy with regular follow-up is appropriate for early detection of subsequent carcinoma [7].

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