

Benign Brenner Tumor Mixed with Mucinous Cystadenoma of the Left Ovary: Case Report and Literature Review, 2023

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Abstract: Ovarian mucinous tumors mixed with Brenner tumors have rarely been reported. The coexistence of such epithelial tumors present histopathologic diagnostic difficulties. Here we report a 57-year-old postmenopausal woman who had experienced an abdominal distention and pain over a period of eight months. A physical examination revealed a grossly distended abdomen that reached the xiphoid process. A firm and mobile abdomino-pelvic mass with a smooth surface and a regular border was identified. Laboratory investigations showed a hemoglobin level of 13.54 g/dl and a serum cancer antigen 125 (CA125) of 97.3 U/mL. Trans abdominal ultrasonography revealed a massive complex mass originating from the left adnexa. A laparotomy was performed and a 10 kg left adnexal mass was removed intact. Histopathological analysis revealed mixed benign mucinous cystadenoma with a Brenner tumor of the left ovary. After surgery the patient showed marked clinical improvement, resumed her regular daily activities in three months and no recurrence has occurred during her long follow up. As the coexistence of these mixed tumors is not uncommon, a thorough pathologic evaluation is necessary and health professionals should be aware of the mixed occurrence of epithelial ovarian tumors.

Keywords: co-existence, Brenner tumor, mucinous cystadenoma, ovarian tumor

Introduction

Ovarian tumors are the most lethal tumors, accounting for 30% of malignancies occurring in the female reproductive tract.¹ It is the leading cause of death from gynecologic malignancy due to its late presentation.¹⁻³ There are still no effective tools for general population screening and it is not cost effective to screen the whole population without risk stratification.⁴ Ovarian cancers are subdivided into type I epithelial ovarian cancers, which are suggested to be relatively indolent and genetically stable tumors that typically arise from recognizable precursor lesions, such as endometriosis or borderline tumors with low malignant potential and type II epithelial ovarian cancers, which are proposed to be biologically aggressive tumors from their outset, with a propensity for metastasis from small volume primary lesions.⁵ Based on origin, primary ovarian tumors are classified into epithelial, stromal and germ cell tumors.¹ Most tumors arise from the surface epithelium and are classified as serous, mucinous, endometrioid, clear cell, and Brenner tumors.^{2,6,7} The World Health Organization (WHO) classifies ovarian neoplasms into 13 distinct categories.^{8,9} A heterogeneous collection of neoplasms, such as mucinous and Brenner tumors, is not uncommon and have appeared in the pathology literature for over a century.^{1,8-11}

Mucinous epithelial ovarian tumors grow to enormous size, making the largest gynecological tumors and representing 10–15% of ovarian neoplasms.^{1,12,13} Usually, at early stages, they are asymptomatic and the patient presents with abdominal swelling and pressure related symptoms.^{2,9}

Histopathologically, mucinous tumors are divided into benign, borderline, and malignant groups. Benign mucinous neoplasms include mucinous cystadenoma and mucinous adenofibroma and account for 80% of cases.⁸ Brenner tumors are relatively rare epithelial neoplasms, accounting for 1.4% to 2.5% of all ovarian tumors. They usually affect postmenopausal women and are mostly benign (99%) in nature and in 30% of benign.^{6,7,14}

The mixed existence of epithelial ovarian tumors is common but the coexistence of mucinous and Brenner tumors is uncommon.^{7,9,14} Minor foci of tumors other than the predominant ones can be ignored, but when substantial volumes (>10%) of several tumor types are present, the tumor is categorized as a mixed tumor.⁸ Mucinous cystadenoma contains a component of a Brenner tumor, in 1.3–4% of ovarian mucinous neoplasms.^{8,14} Interpreting the heterogeneous collection of neoplasms is the most difficult task for surgical pathologists. Due to rarity and disputed histogenesis, Brenner tumors are discussed by pathologists as enigmatic tumors.^{1,14,15} The coexistence of mixed benign Brenner and mucinous tumors should be considered if the imaging finds multilocular or unilocular cystic components with homogeneously hypointense solid components in T2-weighted images.¹¹ Here, we report a 10-kg mixed mucinous cystadenoma and a benign Brenner tumor due to its rarity and diagnostic difficulty in pathology.

Case Report

A 57-year-old para 2 postmenopausal woman visited Orotta National Referral Maternity Hospital on January 18, 2023, complaining of abdominal distension and pain over a period of eight months. Abdominal distention progressed slowly and was associated with decreased appetite and signs of dyspeptic symptoms. She had a history of occasional constipation but no weight loss. Her menses had stopped 10 years back and currently she had no vaginal bleeding or discharge. On physical examination; she was chronically sick looking, with normal vital signs. She had a pink conjunctiva and nonicteric sclera, without cervical or supraclavicular lymph node enlargement. The chest was clear on auscultation, and S1 and S2 were well heard without a murmur or galloping rhythm. The abdomen was grossly distended, reaching the xiphoid process. A large abdomino-pelvic mass measuring approximately 20 cm × 30 cm was identified, firm, mobile, with a smooth surface and regular border. A vaginal examination revealed that the cervix was firm, with a regular edge, and a painless adnexal mass was palpated.

The patient was investigated with locally available investigative modalities and the complete blood count result revealed a white count of 5.20×10^3 , and hemoglobin, hematocrit, and platelet levels of 13.54 g/dl, 44%, and $360.8 \times 10^3/\mu\text{L}$, respectively. On trans-abdominal ultrasonography a huge 30 × 20 × 30 cm complex adnexal mass which is septated originating from the left adnexa was identified. A serum cancer antigen 125 (CA125) level was 97.3 U/mL (normal range: 0–34 U/mL), and carcinoembryonic antigen (CEA), alpha-fetoprotein (AFP), and CA19-9 were within normal ranges.

A laparotomy was performed under general anesthesia and a midline incision was made. A 10 kilograms' massive globular mass, not attached to the anterior abdominal wall or other abdominal organs, originating from the pelvis was identified and extracted without spillage (Figure 1A). Although the uterus, right ovary, and right fallopian tube were healthy, total abdominal hysterectomy with left salpingo-oophorectomy and omentectomy was performed. Intraabdominal lymph nodes were not enlarged, and lymph node dissection was deferred. Macroscopically, the liver, spleen, stomach, and intestine were healthy. After ensuring hemostasis and complete instrument and gauze counts, the abdomen was closed in layers. The extracted mass was sent for histopathological evaluation (Figure 1B), which revealed mixed features of benign mucinous cystadenoma with a Brenner tumor of the ovary (Figure 2). The patient showed dramatic clinical improvement and was discharged with follow-up to the outpatient department to monitor the course of the diseases for possible recurrence. She resumed her regular daily activities in three months and no recurrence occurred during her long follow up.

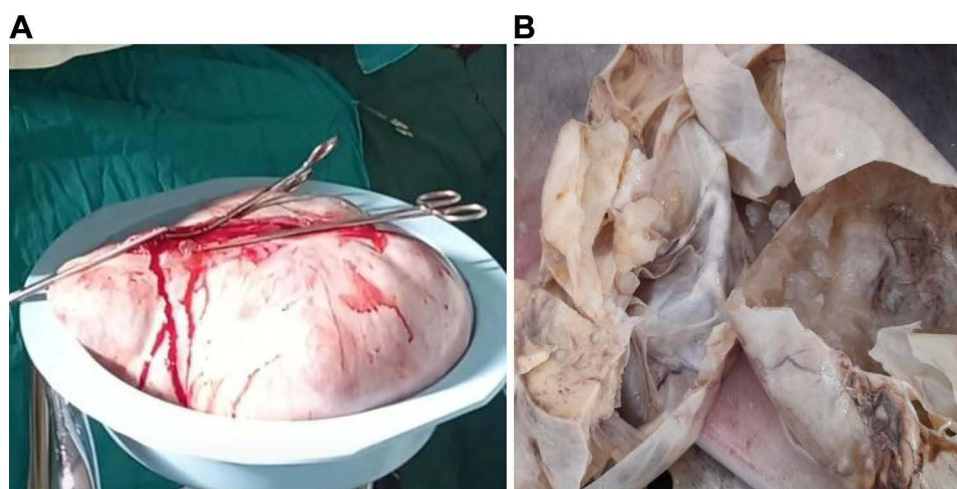


Figure 1 (A) Left ovarian mass 40x30x30 cm, uterus measures 7x5x3 cm, right ovary 2x1x1 cm. (B) The cut section of the cyst show solid area measures 10x7x7 cm after fixation with formalin.

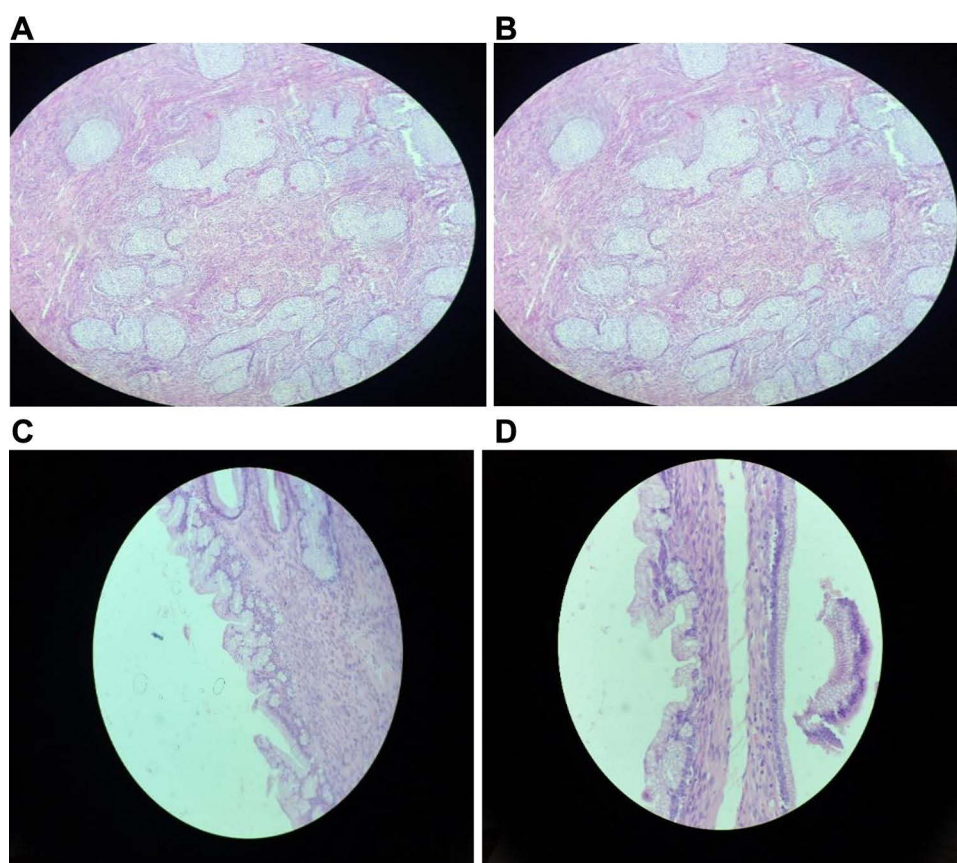


Figure 2 (A and B) Nests of bland looking transitional epithelium within fibromatous stroma showing benign Brenner tumors. (C and D) Section of ovarian cystic lesions showing a multilocular cystic neoplasm composed of multiple cysts and glands lined by a single layer of bland mucinous epithelium showing mucinous cystadenoma.

Discussion

The coexistence of ovarian mucinous tumors with Brenner tumors is uncommon. In such mixed epithelial ovarian tumors the mucinous type consisted the largest part of the tumor and Brenner tumors are frequently identified incidentally.^{9,11} The presence of minor foci of various epithelial tumors can be ignored if it involves (<10%) of the total volume.⁸

A Brenner tumor consists of the solid and cystic nests of epithelial cells resembling a transitional epithelium of the urinary tract and, due to the histologic similarity to the urothelium, formerly Brenner tumors were known as transitional cell tumors.^{3,6,9,15}

Nonspecific symptoms predispose to a late presentation and affects the prognostic outcomes secondary to the associated complications.² In most of the reported cases the course of the disease corresponds to less than 6 months. Occasionally, patients may present with abdominal pain accompanied by vaginal bleeding.^{2,6} Our case presented constitutional symptoms and a change in intestinal habits over a period of 8 months, which is consistent with previous literature. Therefore, patients may not seek medical attention until the appearance of abdominal distension or compressive symptoms. The overall size of such mixed ovarian tumors can be large and reported cases indicate that the majority of them reach >20 centimeters in length.^{8,12} Among ovarian tumours that complicate pregnancies, approximately 5% are malignant. Currently surgical intervention is indicated for an ovarian mass over 6 cm in diameter or when symptomatic.¹⁶ Imaging studying shows a mucinous component frequently presents as a complex cystic multiseptated mass which commonly tends to be larger than simple serous neoplasms.¹¹ Similarly in our case trans-abdominal ultrasonography identified a huge 30 × 20 × 30 cm complex adnexal mass which was septated and originating from the left adnexa.

The intraoperative finding of our case revealed no signs of macroscopic metastasis and lymph nodes were not enlarged. A frozen section was not feasible in our setting which could guide the type of surgery. Omentectomy was performed despite the fact that the omentum was healthy as microcytic metastatic tumors that could change postoperative management could not be ruled out. Appendectomy was also not performed as intraoperative histologic analysis was not performed. Histopathological analysis of coexisting ovarian mucinous with Brenner tumors showed that the size proportion varies and the pathological report of our case revealed that benign mucinous tumors were predominant. Histopathological evaluation revealed that this patient also had endometrial atrophy, which matched ultrasonographic features and the absence of vaginal bleeding.

Conclusion

Coexisting ovarian mucinous with Brenner tumors are uncommon. Imaging finding of multilocular or unilocular cystic components, accompanied by homogeneously hypo-intense solid components should arouse the suspicion of these mixed tumors. A thorough pathological assessment is necessary, and health professionals should be aware of the mixed occurrence of epithelial ovarian tumors.

Data Sharing Statement

All available information is included in the manuscript.

Ethical Approval

The Ministry of Health does not require ethical approval for case reports or case series. Written informed consent was obtained from the patient to share her case details and any accompanying images published globally.

Acknowledgment

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Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors declare that they have no conflict of interest.

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