

Ovarian Brenner Tumor Colliding with Mucinous Cystadenoma: A Case Series

Over Kaynaklı Brenner Tümörü ile Müsinöz Kistadenom Birlikteliği: Bir Olgu Serisi

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Abstract

Ovarian Brenner tumors are rare epithelial neoplasms often found in association with mucinous tumors, particularly mucinous cystadenomas. However, their combined presentation may pose diagnostic challenges due to overlapping clinical and radiological features with those of malignant ovarian neoplasms. We report three cases of ovarian Brenner tumors coexisting with mucinous cystadenomas in postmenopausal women, aged 67-77 years. Clinical manifestations ranged from abdominal distension and pain to incidental findings. Imaging studies consistently demonstrated cystic-solid adnexal masses, sometimes with calcifications or ascites, raising suspicion for malignancy. Histopathological examination confirmed the coexistence of a Brenner tumor and a mucinous cystadenoma in all cases, including one borderline Brenner tumor. Immunohistochemistry supported the diagnosis, with Brenner components expressing urothelial markers such as CK7, p63/p40, and GATA3, while mucinous components displayed distinct immunohistochemical profiles. These cases highlight the importance of recognizing this entity to avoid misdiagnosis and suggest a possible histogenetic relationship between Brenner and mucinous tumors.

Keywords: Brenner tumor, case report, collision tumor, ovarian mucinous cystadenoma

Öz

Over Brenner tümörleri, nadir görülen epitelyal neoplazmlardır ve sıklıkla müsinöz tümörlerle, özellikle müsinöz kistadenomlarla birlikte bulunurlar. Bununla birlikte, bu tümörlerin birlikte görülmesi, malign over neoplazmları ile örtüşen klinik ve radyolojik özellikler nedeniyle tanısal zorluklara yol açabilir. Bu çalışmada, 67-77 yaş aralığında postmenopozal üç kadında Brenner tümörü ile müsinöz kistadenom birlikteliği sunulmaktadır. Klinik bulgular, abdominal distansiyon ve ağrıdan tesadüfi saptamalara kadar değişkenlik göstermiştir. Görüntüleme incelemelerinde, bazen kalsifikasyon veya asit eşlik eden kistik-solid adneksiyal kitleler izlenmiş ve malignite şüphesi doğurmuştur. Histopatolojik inceleme, tüm olgularda Brenner tümörü ile müsinöz kistadenomun birlikte bulunduğunu doğrulamış, bunlardan birinde borderline Brenner tümörü saptanmıştır. İmmünohistokimyasal bulgular tanıyı desteklemiş; Brenner komponentlerinin CK7, p63/p40 ve GATA3 gibi ürotelyal belirteçleri ekspres ettiği, müsinöz komponentlerin ise farklı profiller gösterdiği izlenmiştir. Bu olgular, yanlış tanıyı önlemek açısından bu varlığın tanınmasının önemini vurgulamakta ve Brenner ile müsinöz tümörler arasında olası bir histogenez ilişkisini düşündürmektedir.

Anahtar Kelimeler: Brenner tümörü, olgu sunumu, çarpışma tümörü, over müsinöz kistadenomu



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Introduction

Brenner tumors are uncommon ovarian epithelial neoplasms, usually benign and characterized by transitional-type epithelial nests within dense fibrous stroma, although borderline and malignant variants exist (1,2). They frequently coexist with mucinous ovarian tumors, particularly mucinous cystadenomas, suggesting a possible shared histogenesis. Clinically and radiologically, these lesions often present as cystic-solid adnexal masses, sometimes with calcification, making preoperative diagnosis challenging. We report three cases of ovarian Brenner tumor associated with mucinous cystadenoma and summarize their clinicopathological and imaging features to improve recognition of this diagnostically important entity.

Case Presentation

Case 1

A 77-year-old woman presented with a 4-month history of lower abdominal distension and urinary frequency, without an obvious precipitating cause. She denied any other relevant medical history. Physical examination revealed a palpable mass in the lower abdomen with limited mobility, without tenderness or rebound tenderness. Laboratory tests revealed elevated tumor markers, including carbohydrate antigen (CA) 125 at 53.10 U/mL and CA19-9 at 205.48 U/mL. Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated a giant cystic-solid pelvic mass measuring approximately 17.1×13.2×20.8 cm. The lesion showed heterogeneous density and signal intensity, with multiple calcifications visible within it. No obvious pelvic effusion or abnormally enlarged lymph nodes were identified. Imaging revealed that the pelvic mass was predominantly cystic, with a few internal septa and soft tissue components. On MRI, the lesion showed low signal intensity on

T1-weighted imaging and high signal intensity on T2-weighted imaging (T2WI). Contrast-enhanced imaging demonstrated marked enhancement of the solid components, whereas the cystic components showed no obvious enhancement. Cystadenocarcinoma was initially suspected (Figure 1).

The patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy. Postoperative pathological examination confirmed a borderline Brenner tumor of the left ovary coexisting with a mucinous cystadenoma, with multifocal stromal calcification. Immunohistochemical staining showed that the tumor cells were positive for CK-P, CK7, p63, p53, ER, CA125, and GATA3 and were negative for PR, CK20, CgA, NSE, and S-100. The Ki-67 labeling index was approximately 20%. No recurrence was observed at the 1-year postoperative follow-up.

Case 2

A 71-year-old woman was admitted with a 2-day history of lower abdominal pain. Physical examination revealed lower abdominal distension with mild tenderness, without rebound tenderness. Laboratory tests showed markedly elevated tumor markers, including CA125 at 934 U/mL and CA19-9 at 43.05 U/mL.

Abdominal CT demonstrated a mixed cystic-solid soft-tissue-density mass in the pelvis, with the larger lesion located on the right side and measuring approximately 17.9×12.3×17.0 cm. Contrast-enhanced imaging showed moderately heterogeneous enhancement. The surrounding bowel loops were compressed, and the lesion contained non-enhancing cystic areas and calcifications. In addition, a solid, soft-tissue-density mass measuring approximately 4.6 cm in diameter was identified in the left pelvis; it contained multiple punctate calcifications. Pelvic effusion was also present (Figure 2). The patient underwent a laparoscopic bilateral salpingo-oophorectomy.

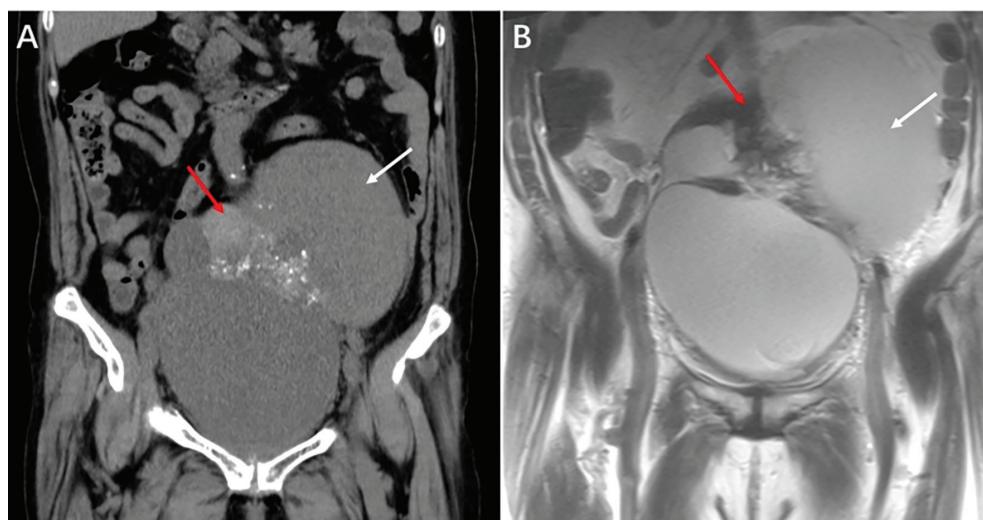


Figure 1. Coronal computed tomography and coronal magnetic resonance imaging demonstrate a large cystic-solid pelvic mass

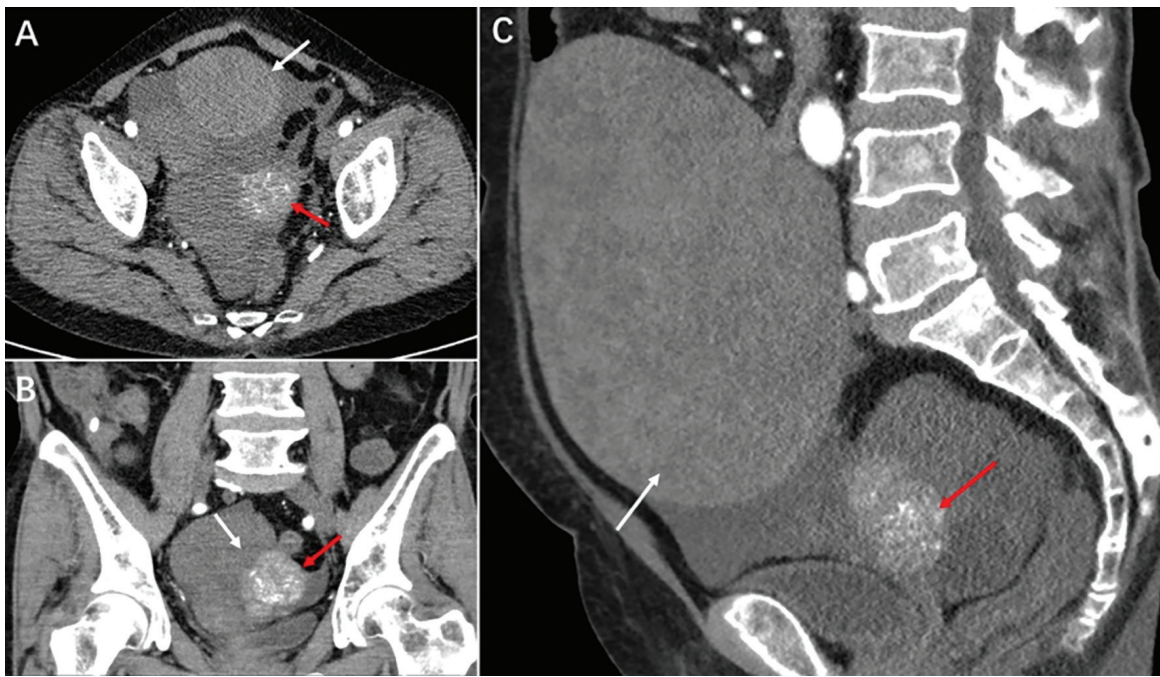


Figure 2. An abdominal computed tomography scan shows a cystic-solid pelvic mass accompanied by pelvic effusion

Postoperative pathological examination revealed a mucinous cystadenoma coexisting with a Brenner tumor in the left ovary. Immunohistochemical and special staining findings were as follows: nests of epithelial-like cells were positive for CK-P, EMA, CK7, WT-1, GATA3, and CA19-9, and negative for CK20, calretinin, and inhibin- α . P53 showed weak-to-moderate positivity in approximately 80% of cells, and the Ki-67 labeling index was 2%. PAS staining was positive. The mucinous epithelium was positive for CKpan, EMA, CK7, and CA19-9, and negative for CK20, calretinin, inhibin- α , WT-1, and GATA3. P53 was partially positive, and the Ki-67 labeling index was 10%. PAS staining was also positive. The patient was lost to follow-up after surgery.

Case 3

A 67-year-old woman was incidentally found to have a pelvic cystic mass on routine ultrasonography 2 months earlier and subsequently presented with dull pain in the left lower abdomen. Her medical history was significant for hypertension for 6 years and for surgery for lung cancer 3 years previously. Laboratory tests showed CA125 of 9.90 U/mL, CA19-9 of 4.52 U/mL, and squamous cell carcinoma antigen of 1.8 ng/mL (elevated). Cervical human papillomavirus testing and cervical cytology showed no obvious abnormalities.

Pelvic MRI demonstrated a round cystic-solid mass in the left adnexal region with well-defined margins, measuring approximately 53×38×46 mm. The lesion showed heterogeneous signal intensity, and part of the cystic fluid exhibited markedly low signal intensity on T2WI. The solid component showed

mildly increased signal intensity on diffusion-weighted imaging and mildly decreased signal intensity on the apparent diffusion coefficient map. Contrast-enhanced imaging demonstrated marked enhancement. A small pelvic effusion was present, and no enlarged lymph nodes were identified. Struma ovarii was initially considered, but an epithelial mucinous tumor could not be excluded (Figure 3).

The patient underwent laparoscopic bilateral salpingo-oophorectomy. Postoperative pathological examination supported the diagnosis of an ovarian Brenner tumor with mucinous metaplasia and calcific deposition, coexisting with an ovarian mucinous cystadenoma with focal epithelial hyperplasia. Immunohistochemical analysis showed that the tumor cells were positive for CK5/6, p40, CK7, CAM5.2, Ki-67 (approximately 25%), p53 (wild-type expression), and GATA3 (partially positive), and were negative for uroplakin 2. No recurrence was observed at the 3-month postoperative follow-up.

Informed consent was obtained from the patient for the anonymous use and publication of clinical and imaging data.

Discussion

Among ovarian tumors, epithelial neoplasms are the most common tumor type and include mucinous and Brenner tumors. Ovarian mucinous tumors account for approximately 10%-15% of all ovarian neoplasms, of which about 80% are benign (1). In contrast, Brenner tumor is a relatively rare epithelial ovarian neoplasm, representing only 1%-2% of all ovarian tumors (2,3).

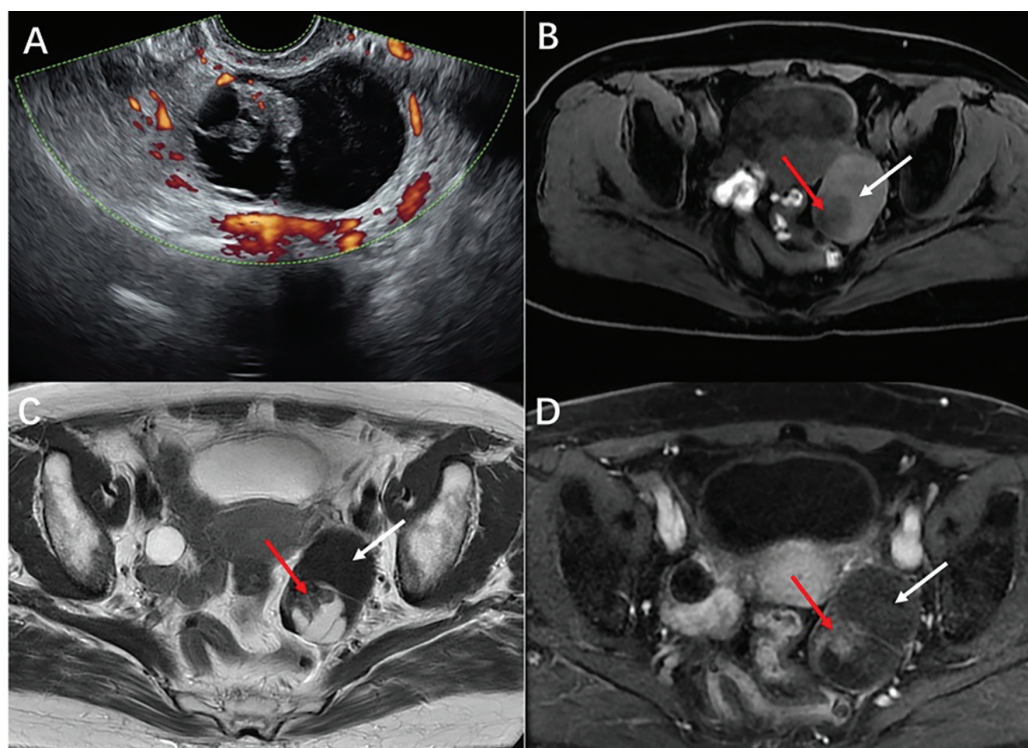


Figure 3. Pelvic ultrasound reveals a cystic-solid mass in the left adnexal region; pelvic magnetic resonance imaging demonstrates a heterogeneous mass in the same region with enhancement of the solid components

It occurs predominantly in postmenopausal women, and the vast majority (approximately 99%) are benign (1).

Brenner tumors are usually small, lack specific clinical manifestations, and are therefore often discovered incidentally during imaging examinations or surgery. In some cases, Brenner tumors may coexist with other types of epithelial ovarian tumors, among which mucinous cystadenoma is the most common. It has been reported that approximately 10% of Brenner tumors are associated with a mucinous tumor component, suggesting a possible biological relationship between these two entities (2). It is currently widely accepted that the coexistence of Brenner tumor and mucinous cystic tumor may arise from a shared origin in the ovarian surface epithelial-stromal complex, whose cells possess multidirectional differentiation potential and are capable of differentiating into different epithelial components (4-6). Some investigators have proposed that the mucinous component may originate from mucinous metaplasia of the Brenner tumor epithelium or that both components may arise from the same precursor lesion. Although the exact pathogenesis has not yet been fully elucidated, molecular pathological studies have identified KRAS mutations in both tumor components in some cases, thereby supporting the hypothesis of a possible clonal origin (7).

From a diagnostic perspective, the mucinous cystic component is often dominant and may obscure the smaller Brenner

tumor component, which typically presents as solid nodules embedded within the cyst wall and can therefore be overlooked. Consequently, careful gross intraoperative examination, systematic postoperative sampling, and meticulous histopathological evaluation are essential for establishing an accurate diagnosis. Although imaging modalities such as ultrasonography and CT are highly sensitive in detecting cystic lesions, their ability to identify small solid nodules is limited, which is a major reason for preoperative or intraoperative misdiagnosis.

From a clinical management perspective, the vast majority of Brenner tumors and mucinous cystadenomas are benign lesions, and complete surgical excision usually results in favorable outcomes. For postmenopausal women, especially when malignancy cannot be completely excluded preoperatively, total hysterectomy with bilateral salpingo-oophorectomy is generally recommended (2). In the present case, the patient was perimenopausal, and the nature of the tumor remained uncertain; therefore, this surgical approach was selected.

Overall, the postoperative prognosis of this type of tumor is favorable. However, it should be noted that both Brenner and mucinous tumors may rarely present as borderline or malignant subtypes, further underscoring the importance of comprehensive histopathological evaluation. In the present case, no obvious malignant features, such as significant cytologic atypia, increased

mitotic activity, or stromal invasion, were identified, thereby confirming that both tumor components were benign.

Conclusion

This case highlights the importance of recognizing composite ovarian tumors, particularly the relatively uncommon but diagnostically significant coexistence of a Brenner tumor and a mucinous cystadenoma. Greater awareness of this entity may help pathologists avoid missed or incorrect diagnoses, while also providing an important basis for appropriate surgical planning and prognostic assessment in clinical practice.

Ethics

Informed Consent: Informed consent was obtained from the patient for the anonymous use and publication of clinical and imaging data.

Footnotes

Authorship Contributions

Concept/Design: W.Y., C.H., Data Collection or Processing: W.Y., Analysis or Interpretation: W.Y., C.H., Literature Review: W.Y., C.H., Writing, Reviewing and Editing: W.Y., C.H.

Conflict of Interest: No conflict of interest was declared by the authors.

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