



AN UNFORESEEN COMBINATION: SIMULTANEOUS PRESENTATION OF BENIGN BRENNERS TUMOR AND BREAST MALIGNANCY IN A SINGLE PATIENT -A RARE CASE REPORT

Radio-Diagnosis

Abishek Deenadayalan*	Junior resident, KS Hegde Medical Academy, Nitte (Deemed to be University), Mangalore, Karnataka, India. *Corresponding Author
Nireeksha Shetty	Assistant professor, KS Hegde Medical Academy, Nitte (Deemed to be University), Mangalore, Karnataka, India.
Varun Pallamala	Junior resident, KS Hegde Medical Academy, Nitte (Deemed to be University), Mangalore, Karnataka, India.

ABSTRACT

Brenner tumors are rare epithelial tumors that account for around 2% of ovarian neoplasms. Computed tomography features of Brenner tumor are specific for diagnosis which includes the presence of calcifications, cystic component with septae, and enhancing solid component. Brenner ovarian tumor in patients with breast malignancy is rare. We present a rare case of benign ovarian Brenner tumor in an elderly woman diagnosed with invasive ductal carcinoma of breast.

KEYWORDS

Brenner tumor, Ovarian neoplasms, Breast malignancy

INTRODUCTION

Brenner tumors (BT) are rare epithelial tumors that account for around 2% of ovarian neoplasms. (1) BT are classed as benign, borderline, or malignant by the World Health Organisation. Borderline and malignant variants are extremely rare, accounting for 10% of BT cases and having a higher age range.

The average age at presentation is 50 years with 71% of the patients being more than 40 years. It is generally asymptomatic and most often incidentally discovered on imaging. (2). Histologically, they are constituted of nests of urothelial-like epithelial cells surrounded by a dense fibrous stroma. Calcific foci within the stroma have been identified in around 50% of cases. (3) To our knowledge, the association between Brenner tumor of the ovary and invasive ductal carcinoma of breast is extremely rare. In the present report, we describe a rare case of benign ovarian Brenner tumor in an elderly woman diagnosed with invasive ductal carcinoma of breast.

Case Presentation:

Clinical details:

A 59 years old female, who is a known case of invasive ductal breast carcinoma, presented with the complaints of abdominal pain and abdominal distension for about 15 days. On per abdomen examination there was a firm, non-tender mass measuring 15 x 20 cm in right iliac and lumbar regions extending up to the umbilical region.

Imaging findings:

Ultrasound abdomen and pelvis revealed a large solid cystic lesion arising from the right adnexa extending from pelvis to the epigastrium. The lesion showed multiple thin and thick internal septations. Both ovaries were not separately visualized from the lesion.

Moderate free fluid was seen within the peritoneal cavity. Based on the sonographic findings, a diagnosis of complex abdominopelvic mass, likely of ovarian origin was made and CECT (Contrast-enhanced Computed Tomography) of abdomen and pelvis with thorax cuts was advised for further evaluation. Unenhanced CT (Computed Tomography) images showed a well-defined mixed density lesion with calcifications within. (Figure 1) CECT images revealed a well-defined lobulated heterogeneously enhancing lesion measuring 15.3 x 18 x 19 cm (ap x tr x cc) in the abdominopelvic region in the midline. The lesion showed multiple non enhancing necrotic areas with multiple enhancing septae and an enhancing solid component within. (Figure 2)

Anteriorly, the lesion was abutting the anterior abdominal wall muscles with obliterated fat planes, and posteriorly, the lesion was abutting the aorta and iliac vessels (Figure 3 A). Superiorly, the lesion was

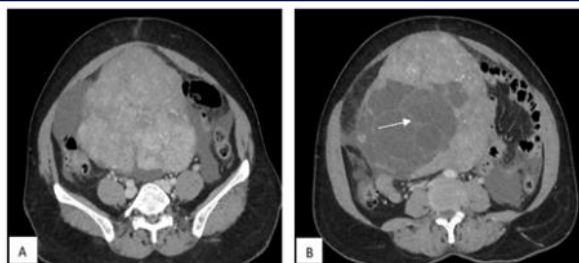


Figure 1. An unenhanced CT axial image shows a well-defined mixed density lesion with calcifications within (White arrows).

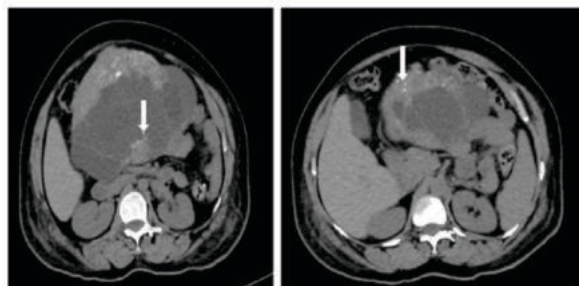


Figure 2. CECT axial images in venous phase shows heterogeneously enhancing lesion with solid (A) and cystic (B) components and internal enhancing septae (White arrow).

extending to the level of supra-umbilical region and displacing the bowel loops, and inferiorly, the lesion was extending up to the superior surface of the urinary bladder with indistinct fat planes. (Figure 3 B)

The vascular supply of the lesion was seen arising from the right gonadal and uterine arteries (Figure 4).

Based on the imaging features, the differential diagnosis of BT, fibrothecoma and malignant ovarian neoplasm were given.

CT sections of the thorax showed a well-defined lobulated hypodense lesion in the upper inner quadrant of left breast. The lesion showed spiculated margins with perilesional architectural distortion. Multiple enlarged discrete and conglomerated left axillary lymph nodes were seen (Figure 6).

Histopathology:

The tissue from the abdominopelvic mass was composed of fibrocollagenous ovarian stroma and benign islands of transitional cells (Figure 5). No definite evidence of malignancy was detected. Based on this tissue sampling, the final diagnosis of Benign BT was made.

DISCUSSION:

BT is an unusual epithelial-stromal tumor that arises from the surface of the ovaries (4) and was first described by Fritz Brenner in 1907. (5) BT has epithelial cell nests developing in fibrous stroma histologically. (6) Benign BT can occur in women of any age; however, they are most common in women over the age of 50, whereas women with borderline or malignant BT are usually 10 years older. (7) The majority of BT documented in the literature are discovered incidentally or as a result of symptoms due to mass effect. Common manifestations in symptomatic patients include vaginal bleeding, a palpable pelvic mass, and pelvic pain. (8)

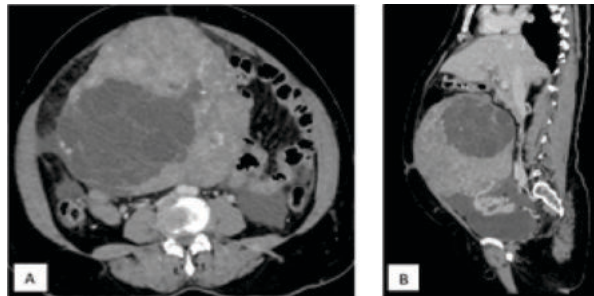


Figure 3: CECT images in venous phase - axial (A) and sagittal (B) sections. Anteriorly, the lesion is abutting the abdominal wall muscles. Posteriorly the lesion is abutting the iliac arteries. Superiorly, the lesion is extending to the supra-umbilical region. Inferiorly, the lesion is extending up to the superior surface of urinary bladder.

The lesion of interest in our patient is a well-defined lobulated heterogeneously enhancing lesion in the abdominopelvic region in the midline. The lesion shows multiple non-enhancing necrotic areas, multiple enhancing septae, a solid enhancing component and multiple punctate calcific foci within. In a study conducted by P.-F. Montoriol et al. (2) benign BT were unilateral in 89% of patients, with 49% solid and 51% mixed solid cystic lesions. Calcifications were seen on CT in 70.4% of lesions that were punctate and scattered. The findings in our case are consistent with the study by P.-F. Montoriol et al. (2) Calcifications have been reported in 83% of BT on CT in a study conducted by Green GE et al. (8) and most of the lesions were solid, whereas in our case, the lesion is mixed solid cystic.

BT is frequently associated with other ovarian tumors, which share a common origin from the coelomic epithelium and are all estrogen dependent. Only a few of these tumors have been associated to other extra-ovarian tumors. (9) The presence of a BT in a patient with known breast malignancy is rare and this association is not yet established. Indraccolo U et al. (10) presented a case of bilateral BT with endometrial adenocarcinoma in a postmenopausal woman. Belloni AR et al. (11) reported an unusual association of a BT and adenocarcinoma of the sigmoid colon.

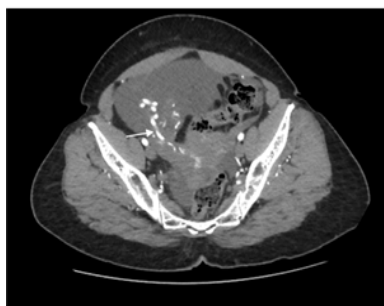


Figure 4: Arterial phase CT axial image demonstrating the vascular supply of the lesion (White arrow) arising from right gonadal artery.

The differential diagnosis of BT includes cystadenocarcinoma, fibroma, leiomyoma and Krukenberg tumor. (12) Cystadenocarcinoma presents as an adnexal cystic solid mass with an unclear border and cystic-solid borderline, an unequally thickened cystic wall with multiple nodules and septa, and is usually accompanied by peritoneal metastasis. (13) Fibroma often manifests as a homogenous solid mass with a smooth border and minimal enhancement. Cystic changes, calcifications and necrosis can be seen occasionally. (14) Leiomyomas present with a limited amount of dot or eggshell-like calcification with a similar enhancement pattern as BT.

(15) Krukenberg tumors are typically bilateral, with additional primary malignancy characteristics, and are frequently associated with ascites or metastases. (16)

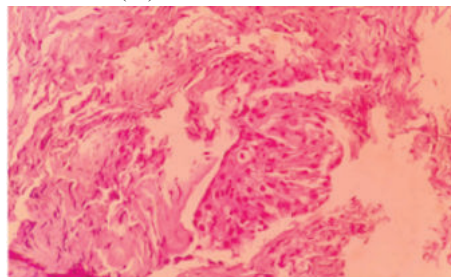


Figure 5: Histopathological examination image shows core tissue with fibrocollagenous ovarian stroma and benign islands of transitional cells.

In the present case, the typical imaging features of BT helped us in the making the diagnosis. However, a differential diagnosis of metastasis was considered because of the presence of the primary invasive ductal carcinoma of the breast. Diagnosis of BT was confirmed on histopathology. Patient was advised 3 cycles of neoadjuvant chemotherapy, followed by modified radical mastectomy and laparotomy for ovarian tumor.

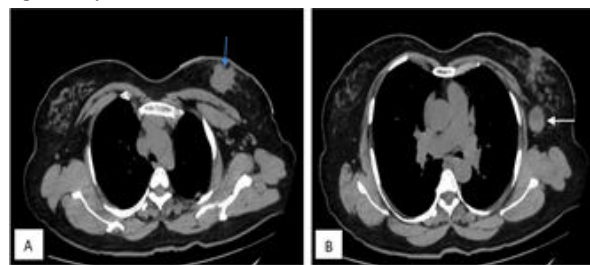


Figure 6: Axial CT sections of the thorax showing - A: A well-defined lobulated hypodense lesion in the upper inner quadrant of left breast (Blue arrow). The lesion shows spiculated margins. B: Enlarged left axillary lymph node (White arrow).

CONCLUSION:

An ovarian lesion in a patient with a known breast malignancy is expected to be a metastatic lesion. However, there is a possibility of that lesion to be benign as described in our case. BT has characteristic and pathognomonic CT features, like whorled appearance, calcification, necrosis and solid component. Brenner ovarian tumor detected in patients with known breast malignancy is rare and is likely a result of two separate events than an association. Hence, it is important to know the imaging features of primary ovarian neoplasms, more importantly the benign tumors to differentiate them from the secondary lesions.

Abbreviations:

BT - Brenner tumors

CECT – Contrast enhanced Computed Tomography

CT - Computed Tomography

REFERENCES:

1. Outwater EK, Siegelman ES, Kim B, Chiowanch P, Blasbalg R, Kilger A. Ovarian Brenner tumors: MR imaging characteristics. *Magnetic resonance imaging*. 1998 Dec 1;16(10):1147-53.
2. Montoriol PF, Hordonneau C, Boudinaud C, Molnar I, Abrial C, Kossai M. Benign Brenner tumour of the ovary: CT and MRI features. *Clin Radiol*. 2021 Aug;76(8):593-598.
3. Vemavarapu L, Alatassi H, Moghadamfalahi M. Unusual presentation of benign cystic Brenner tumor with exuberant psammomatous calcifications. *Int J Surg Pathol* 2011;19(1):120e2.
4. Dierickx I, Valentin L, Van Holsbeke C, Jacomen G, Lissoni AA, Licameli A, Testa A, Bourne T and Timmerman D: Imaging in gynecological disease (7): Clinical and ultrasound features of Brenner tumors of the ovary. *Ultrasound Obstet Gynecol* 40: 706-713, 2012.
5. Balasa RW, Adcock LL, Prem KA and Dehner LP: The Brenner tumor: A clinicopathologic review. *Obstet Gynecol* 50: 120-121, 1977.
6. Ordóñez NG and MacKay B: Brenner tumor of the ovary: A comparative immunohistochemical and ultrastructural study with transitional cell carcinoma of the bladder. *Ultrastruct Pathol* 24: 157-167, 2000.
7. Silverberg SG: Brenner tumor of the ovary. A clinicopathologic study of 60 tumors in 54 women. *Cancer* 28: 588-596, 1971.
8. Green GE, Morteel KJ, Glickman JN, Benson CB. Brenner tumors of the ovary sonographic and computed tomographic imaging features. *J Ultrasound Med* 2006;25:1245-51

9. Ziadi S, Trimeche M, Hammadi F, Hidar S, Sriha B, Mestiri S, et al. Bilateral proliferating Brenner tumor of the ovary associated with recurrent urothelial carcinoma of the urinary bladder. *North Am J Med Sci.* 2010;2:39–41.
10. Indraccolo U, Cingolani N, Indraccolo SR. Bilateral Brenner tumor with endometrial adenocarcinoma in a postmenopausal woman. *Eur J Gynaecol Oncol* 2007; 28: 233-234.
11. Belloni AR, Germani G. Neoplastic associations of Brenner tumor. A propos of a case associated with adenocarcinoma of the sigmoid. *Pathologica* 1990; 82: 101-108.
12. Zhao Y, Mao X, Yao L, Shen J. Computed tomography imaging features of benign ovarian Brenner tumors. *Oncol Lett.* 2018;16(1):1141-1146. doi:10.3892/ol.2018.876
13. Moon WJ, Koh BH, Kim SK, Kim YS, Rhim HC, Cho OK, ahm CK, Byun JY, Cho KS and Kim SH: Brenner tumor of the ovary: CT and MR findings. *J Comput Assisted Tomogr* 24; 72-76, 2000.
14. Jung SE, Lee JM, Rha SE, Byun JY, Jung JI and Hahn ST: CT and MR imaging of ovarian tumors with emphasis on differential diagnosis. *Radiographics* 22: 1305-1325, 2002.
15. Bristow R and Montz F: Leiomyomatosis peritonealis disseminata and ovarian Brenner tumor associated with tamoxifen use. *Int J Gynecol Cancer* 11: 312-315, 2001.
16. Imaoka I, Wada A, Kaji Y, Hayashi T, Hayashi M, Matsuo M and Sugimura K: Developing an MR imaging strategy for diagnosis of ovarian masses. *Radiographics* 26: 1431-1448, 2006.