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Case Report

A rare presentation of benign Brenner tumor of ovary: a case report

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ABSTRACT

Brenner tumors are rare ovarian tumors accounting for 2-3% of all ovarian neoplasms and about 2% of these tumors are borderline (proliferating) or malignant. These tumors are commonly seen in 4th-8th decades of life with a peak in late 40s and early 50s. Benign Brenner tumors are usually small, <2cm in diameter and often detected incidentally during surgery or on pathological examination. Authors report a case of a large, calcified benign Brenner tumor in a 55-year-old postmenopausal woman who presented with complaint of abdominal pain and mass in abdomen. Imaging revealed large complex solid cystic pelvic mass -peritoneal fibrosarcoma. She underwent laparotomy which revealed huge Brenner tumor weighing 9kg arising from left uterine cornual end extending up to epigastric region. Myomectomy and hysterectomy with bilateral salphingo oophorectomy, omentectomy done and the specimen was sent for histopathological and immunohistochemistry study which revealed benign Brenner tumor. Brenner tumors are rare ovarian neoplasms accounting for 2-3% of all ovarian tumors. Benign Brenner tumors are usually small and solid whereas borderline and malignant Brenner tumors are usually larger and cystic with solid areas. But it is possible to have a completely benign large Brenner tumor. Therefore, benign nature of the lesion should not be excluded even when the ovarian tumor is very large.

Keywords: Benign, Borderline, Malignant Brenner tumors, Proliferating

INTRODUCTION

Brenner tumors are uncommon solid fibroepithelial tumor resembling fibroma of ovary on gross appearance.¹ They are rare ovarian tumors constituting between 2-3% of all ovarian neoplasms.

About 6-7% is bilateral. Most of these tumors are benign, <2cm in size and usually located in the cortex. Sometimes they may present as a mural nodule in a mucinous cystadenoma, mature cystic teratoma or rarely struma ovarii.² They may also be seen in association with transitional cell tumors of urinary bladder.³

Borderline and malignant Brenner tumors are usually larger than benign Brenner tumors. Brenner tumors are

transitional cell tumors of the ovary which include benign, borderline (proliferating, proliferative) and malignant varieties.⁴ Some patients with Brenner tumors may show signs of hyper estrogenism. 10-15% cases show mild endometrial hyperplasia and the patient may present with menorrhagia or postmenopausal bleeding.

Occasionally it may be associated with hydrothorax or ascites (pseudomeigs syndrome). In rare cases it may become malignant.

These tumors are generally seen in postmenopausal women and is generally unilateral, small to moderate in size, essentially benign and has no endocrine function and cut surface appears gritty and yellowish grey.

CASE REPORT

A 55-year-old female came to surgery OPD with C/O mass per abdomen for 2 months. C/O upper abdominal pain, dull aching, not radiating, no specific aggravating and relieving factors. She had H/O loss of appetite, loss of weight, H/o dyspepsia, H/O constipation. Patient is a known hypertensive for 2 years on treatment. Menstrual H/O: Regular menstrual cycles. Attained menopause at 48 years. Patient was diagnosed as a case of peritoneal fibrosarcoma and investigated O/E. She was conscious, oriented, afebrile, hydration fair, no pallor, CVS-S1S2+, RS-BAE+, P/A-Umbilicus pushed towards right, mass palpated in epigastric, left and right paraumbilical region, hard in consistency, not freely mobile, abdominal distension+. On knee elbow position mass falls forward. P/R: normal. MRI pelvis showed lobulated mass lesion with calcification probably peritoneal fibrosarcoma. small fibroid uterus. CECT abdomen showed 24 X 21cm irregular lobulated mass lesion with calcification and necrosis is seen adjacent to uterus.

Features suggestive of leiomyosarcoma / Mullerian malignancy. CECT chest – normal study. Her CA 125 was 39.5U/mL. Under general anaesthesia patient in supine abdomen opened by midline transverse incision, huge mass of weight 9 kg excised (Figure 1), mass found to be arising from left uterine cornual end feeding vessel for the mass was left uterine artery. About 50 ml of ascitic fluid let out. Hysterectomy, salpingo oophorectomy with omentectomy done. Wound closed in layers with perfect hemostasis.



Figure 1: Large mass with bosselated surface weighing 9 Kg.

Postoperative period was uneventful. From macroscopic examination authors considered the possibility of a large sub serous fibroid with adherent ovary and fallopian tube showing extensive calcification. Cut surface was firm to hard, grey white, solid with extensive areas of calcification (Figure 2) microscopic examination showed. Section studied from uterus showed atrophic endometrium,

myometrium showed neoplasm composed of fascicles of spindle cells.

Cervix shows features of chronic cervicitis. Right ovary and fallopian tube showed normal histology. Section from omentum shows congestion, ascitic fluid showed chronic and mesothelial cells in an eosinophilic fluid background, no malignant cells. Section showed from left ovary neoplasm composed of nests of transitional cells embedded in a fibrous stroma. Foci of calcification seen. IHC panel revealed Cytokeretin- Positive, Cytokeretin 20- Negative, Vimentin& Smooth muscle actin - Negative for epithelial component, Positive for spindle cells, Inhibin-Negative, CA 125, WT1- Negative, KI-67- 01% Positive, Calretinin -Negative. With this authors made the diagnosis of benign Brenner tumor of leftovary.

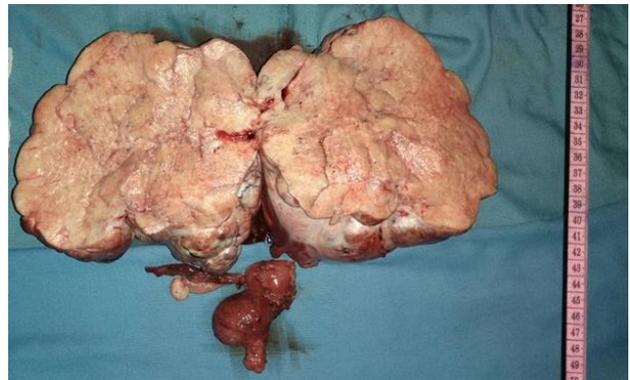


Figure 2: Firm to hard, grey white, solid with extensive areas of calcification.



Figure 3: Section showed from left ovary neoplasm composed of nests of transitional cells embedded in a fibrous stroma. Foci of calcification seen.

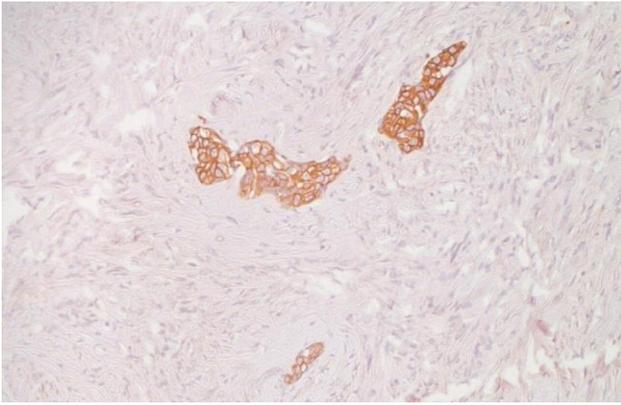


Figure 4: Immunohistochemistry positive for cytokeratin-7.

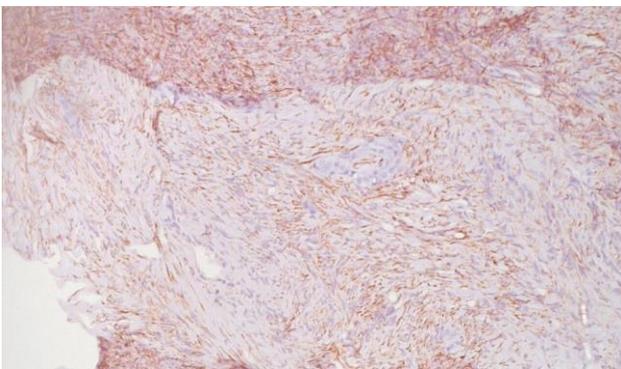


Figure 5: Immunohistochemistry positive for vimentin.

DISCUSSION

Brenner tumor of ovary is a solid ovarian tumor that is generally asymptomatic. Although they are predominantly solid on imaging and pathologic examination, association with serous and mucinous cystadenomas is up to 30%.⁵ It is usually an incidental pathological finding. Among symptomatic patients, common symptoms include vaginal bleeding, a palpable pelvic mass, and pelvic pain. Most of the time it is found to be unilateral. Bilaterality is seen only in 5–7% of the cases.⁶ It is generally accepted that Brenner tumors are derived from the surface epithelium of the ovary or the pelvic mesothelium through transitional cell metaplasia to form the typical urothelial-like components.⁷ The histological patterns observed in Brenner tumor are typically benign, with a few reports of borderline or malignant counterparts.⁸ Majority of them are benign, <2% being borderline or malignant. Pure transitional cell carcinoma of ovary is very rare (1%) of surface epithelial carcinomas of ovary.

Benign Brenner tumors are unilateral, small (<2cm) and solid. They are usually well circumscribed with a firm grey white or yellowish white cut surface. It closely resembles of a fibroma or thecoma. Occasionally benign tumors may be partly cystic, and some tumors may be

hard and gritty due to calcification. It is difficult to diagnose Brenner tumor with imaging studies. USG and computed tomography, both the techniques are limited in specificity because of the tumor's nonspecific appearance. In imaging studies benign Brenner tumors are generally similar to those of other solid ovarian masses such as fibroma, fibrothecoma, and pedunculated leiomyoma. Nuclear grooves may be present, stroma may show focal hyalinization and calcific plaques. 10-15% cases show luteinized cells in the stroma and are associated with features of hyperestrogenism. Brenner tumors can be seen in association with mucinous cyst adenoma, struma ovarii or strumal carcinoid. Benign Brenner tumor with large mucinous cyst may be misdiagnosed as mucinous cyst adenoma unless transitional cells at the periphery of the mucinous cells are identified.

Immuno Histo Chemistry: The cells show cytoplasmic positivity for keratin, EMA or CEA. Scattered argyrophilic cells positive for chromogranin and 5HT may be present. Borderline or proliferating or proliferative Brenner tumors present as unilateral. They are larger than benign (8-30cm) in diameter and partly cystic with polyps and friable papillae. Projecting into the lumen, Papillary structures are lined by transitional epithelium. Lining cells resemble grade 1 to grade 3 papillary urothelial carcinoma of urinary tract with no invasion into the stroma. A benign Brenner component is also present. Mucinous cells may be present. Borderline Brenner tumors composed of grade 1 malignant urothelial cells are designated as borderline tumors (not otherwise specified) and tumors with grade 2 or 3 malignant urothelial cells are regarded as borderline tumors with intra epithelial carcinoma.^{9,10} There are only a few cases of borderline Brenner tumors which produced local recurrence or metastasis, all other reported lesions pursued a relatively benign course. So unilateral salpingo oophorectomy is considered adequate treatment for these tumors.

Grossly benign Brenner tumors are well circumscribed, with a hard or fibromatous, gray, white, or slightly yellow cut surface. Occasionally the tissue becomes gritty because of calcific deposit. Borderline Brenner tumors are characteristically cystic and unilocular or multilocular with cauliflower like papillomatous masses protruding into one or more of the locules. Malignant Brenner tumor may be solid or cystic with mural nodules; they usually do not have any distinctive features.¹¹

Microscopically, they are made of abundant dense fibrous stroma with epithelial nests of transitional cells resembling those lining the urinary bladder. The fibrous component is less prominent in borderline or malignant tumors than in benign lesions. Complex cystic tumors contain varying amounts of stroma and are more commonly found with borderline or malignant histologic findings, often in the form of papillary solid projections within a cystic mass.¹² Malignant Brenner tumors are

usually unilateral, 5-25 cm in diameter and predominantly cystic with solid areas.

Microscopy shows grade 1 to grade 3 nests of urothelial carcinoma or focal squamous cell carcinoma irregularly infiltrating into the stroma. Mucinous component may be present. According to WHO recommendation irregular infiltration into the stroma is the most important diagnostic criteria for malignant Brenner tumor. Borderline and malignant Brenner tumors may be difficult to be distinguished from metastatic urothelial carcinoma from urinary bladder. A careful search for benign Brenner tumor nests or mucinous cells should be done to rule out the possibility of metastasis. Pure form of transitional cell carcinoma (TCC) accounts for only 1% of surface epithelial carcinomas. A carcinoma of transitional type not associated with benign Brenner tumor is called a transitional cell carcinoma of ovary. Primary TCC may mimic metastatic TCC from urinary bladder. In difficult cases immunohistochemistry will aid in the diagnosis.

Urinary tract TCC cells are reactive for CK 20 and thrombomodulin in contrast to primary ovarian TCC. It is difficult to differentiate primary high grade TCC of ovary from other poorly differentiated surface epithelial carcinomas. TCC is more aggressive but responds to chemotherapy better and has got a higher survival rate compared to other high grade advanced ovarian cancers. In present case also the radiologist considered the possibility of a large sub-serous fibroid following ultrasound and CT scan studies. Generally benign Brenner tumors are small lesions usually <10cm in diameter. Larger size of the tumor is suggestive of malignant potential of the tumor. However, it is possible to have a completely benign, large Brenner tumor. Therefore, authors cannot rule out the possibility of a benign Brenner tumor even when the tumor is very big.

CONCLUSION

Brenner tumors are rare ovarian neoplasms accounting for 2-3% of all ovarian tumors. Benign Brenner tumors are usually small and solid whereas borderline and malignant Brenner tumors are usually larger and cystic with solid areas. But it is possible to have a completely benign large Brenner tumor. Therefore, benign nature of the lesion should not be excluded even when the ovarian tumor is very large.

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