Journal of Cancer Research and Practice

journal homepage: www.ejcrp.org

Case Report

Brenner Tumor of the Ovary: Incidental Finding with Unusual Combinations

Hema Udawat[1], Vandana Nunia[2*], Natasha Agarwal[1], Arpita Jindal[1]

[1]Department of Pathology, SMS Medical College, Jaipur, Rajasthan, India
[2]Department of Zoology, University of Rajasthan, Jaipur, Rajasthan, India

Abstract

The most common ovarian tumors are surface epithelial tumors, comprising 58% of all ovarian tumors. Among them, serous and mucinous cystadenomas are the most common (35%). A Brenner tumor is a rare, mostly benign type of ovarian epithelial neoplasm with an incidence of 1%–2%. Rarely, they occur in other locations, including the testis. The majority of Brenner tumors are benign; however, some can be malignant. These are most frequently found incidentally on pelvic examinations or during laparotomy. In the present case series, we report three cases of Brenner tumors diagnosed incidentally on histopathology. The first case was a rare combination of serous cystadenoma, with a focus showing a Brenner tumor in the wall of serous cystadenoma. The second case was a proliferating Brenner tumor in the wall of mucinous cystadenoma, and the third case had bilateral benign Brenner tumors.

Keywords: Brenner tumor, mixed surface epithelial tumor ovary, serous cystadenoma

Introduction

Surface epithelial tumors are the most common type of ovarian tumors,[1] occurring at all ages with a peak incidence in the second to fifth decades of life. Various combinations of these epithelial tumors have been described.[2] A Brenner tumor of the ovary is a solid ovarian tumor that is generally asymptomatic. Although they are predominantly solid on imaging and pathologic examinations, up to 30% of cases are associated with serous and mucinous cystadenomas.[3] Such tumors are usually an incidental pathological finding. Most cases are unilateral, and bilateral Brenner tumors are seen in only 5%–7% of cases.[4] 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.[5] The histological patterns observed in Brenner tumors are typically benign, with a few reports of borderline or malignant counterparts.[6] Malignant cases are extremely rare (2%), and so are proliferative Brenner tumors. A serous cystadenoma coexisting with a benign Brenner tumor is also rare.[7-9]

It is difficult to diagnose a Brenner tumor with imaging studies. Ultrasonography and computed tomography are limited in

Received: 11-Jun-2018 **Revised:** 04-Sep-2018 **Accepted:** 12-Sep-2018

Access this article online

Quick Response Code:

Website: www.ejcrp.org

DOI: 10.4103/JCRP.JCRP_3_18

Address for correspondence: Dr. Vandana Nunia, Department of Zoology, University of Rajasthan, Jaipur - 302 004, Rajasthan, India.
E-mail: vandananunia@gmail.com

How to cite this article: Udawat H, Nunia V, Agarwal N, Jindal A. Brenner tumor of the ovary: Incidental finding with unusual combinations. J Cancer Res Pract 2019;6:33-6.

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.

In this study, we report three cases of Brenner tumors, all of which were diagnosed incidentally on pathological examinations. This study was approved by our Institutional Ethics Committee (No 528/MC/EC/2013, dated 15.02.2013).

Case Reports

Case 1

A 50-year-old female patient presented with lower abdominal pain, bloating, and an obvious gradually increasing swelling for the past 6 months. She had no comorbid illnesses and she had not previously undergone surgery. Biochemical and hematological profiles were normal. Ultrasonography of her abdomen revealed a cystic ovarian mass on the left side. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed, and the specimen was sent for histopathological examination. The cut-open specimen of the uterus and cervix measured 6 cm × 4 cm × 3 cm. An intramural fibroid measuring 0.4 cm was seen on serial cut sections. The left ovary was cystic and measured 7 cm in diameter, and the external surface was smooth with prominent blood vessels. A clear fluid was released on cutting, and the internal surface was rugose. The cyst wall was 0.2 cm thick, and no papillary excrescence or solid areas were seen. The right ovary measured 3 cm × 2 cm × 1 cm, and the external and internal surfaces were unremarkable [Figure 1]. Microscopy revealed a leiomyoma in the uterus. The cervix, both fallopian tubes, and right ovary were normal. The left ovarian cyst wall was lined by cuboidal-to-columnar epithelium, and at places was thrown into papillary projections. At one focus, the wall of the cyst showed a nest of transitional epithelial cells surrounded by fibrous stroma. The cells contained a moderate amount of eosinophilic cytoplasm and round-to-oval grooved nuclei. A diagnosis of serous cystadenoma with a Brenner tumor was made [Figure 2a].

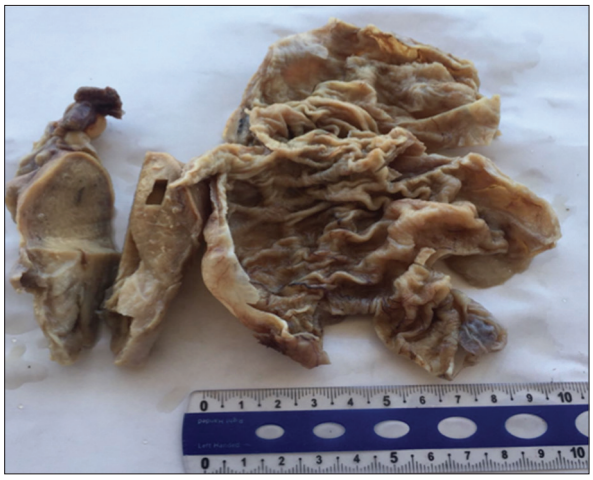

Figure 1: Gross specimen of hysterectomy with bilateral salpingo-oophorectomy showing an ovarian cyst of 7 cm size (left side) with an internal wall showing mucosal rugosities

Case 2

A 40-year-old female patient presented with lower abdominal pain and an obvious gradually increasing swelling for the past 2 months. She had no comorbid illnesses and had never undergone surgery. Hematological and biochemical profiles were normal. Ultrasonography of her abdomen revealed a large cystic mass with multiple internal septations, extending from the pelvic cavity to high up in the epigastrium, more on the right side. Fine internal echoes were seen, but no calcification was noted. A mucinous cystadenoma was suspected clinically. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed, and the specimen was sent for histopathological examination. The cut-open specimen of the uterus and cervix measured 6 cm × 3 cm × 3 cm. Both fallopian tubes were unremarkable. The left ovary measured 3 cm × 2 cm × 1 cm, and the cut surface showed a cyst of 1 cm in size. The right ovary had been converted into a cyst measuring 14 cm × 13 cm × 13 cm, which was multilocular and filled with mucinous material. The walls were papery thin to 1 cm thick. There was a gray white-to-yellow firm area measuring 3.5 cm × 3.5 cm × 3 cm which appeared to be encapsulated. Microscopically, the uterus, cervix, and both tubes were unremarkable. The left ovary showed foci of a benign Brenner tumor, and the right ovarian cyst showed features of mucinous cystadenoma with foci of a Brenner tumor in the cyst wall. A diagnosis of mucinous cystadenoma on the right side with bilateral Brenner tumors was made [Figure 2b].

Case 3

A 60-year-old female presented with lower abdominal pain for the last 6 months. She had no comorbid illnesses and had not previously undergone surgery. Biochemical and hematological profiles were normal. Ultrasonography of her

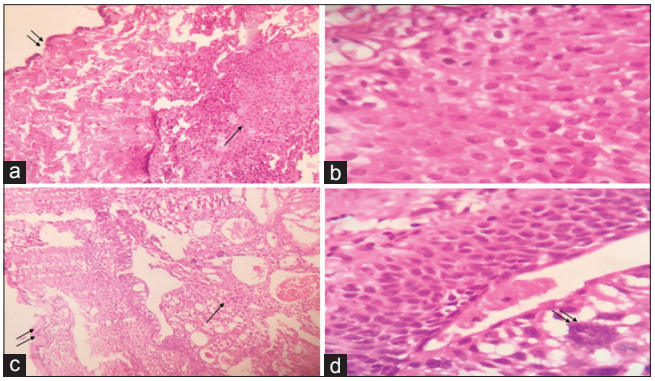

Figure 2: Microscopic description of the Brenner tumor: (a) H and E stained section showing a cyst lined by cuboidal-to-low columnar epithelium (double arrow) and nests of Brenner tumors in the wall of the cyst (single arrow) (×100). (b) H and E-stained section showing nests of Brenner tumors. The cells had a moderate amount of eosinophilic cytoplasm and grooved nuclei (×400). (c) H and E-stained section showing the cyst wall lined by tall columnar mucinous epithelium (double arrow) and a proliferating Brenner tumor in the wall of the cyst showing atypical nuclear features with no stromal invasion (single arrow) (×100). (d) H and E-stained section showing a proliferating Brenner tumor with moderate nuclear atypia (double arrow) (×400)

abdomen revealed a cystic mass in the left pelvic region. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was done and sent for histopathological examination. The cut-open specimen of the uterus and cervix measured 5 cm × 3 cm × 2 cm. Both fallopian tubes and left ovary were unremarkable. The right ovary had been converted into a cyst of 10 cm × 10 cm × 8 cm, and the external surface was smooth. A watery fluid was released on cutting. The internal surface showed prominent vascular markings, and the wall thickness was 0.1–0.2 cm. Microscopically, the cervix, both fallopian tubes, and the left ovary were unremarkable. Endometrium showed cystic atrophy. The right ovarian cyst showed features of mucinous cystadenoma, and the walls showed foci of a Brenner tumor. A single focus showed atypical features without stromal invasion. A diagnosis of mucinous cystadenoma with a proliferating Brenner tumor was made [Figure 2c and d].

Discussion

Brenner tumors are most commonly associated with mucinous cystadenomas. Other associations include germinal inclusion cysts, simple cysts, struma ovarii, cystic teratomas, stromal sarcomas, and chocolate cysts of the ovary.[10-12] Mucinous cystadenomas of the ovary occasionally contain small nodules of Brenner tumor. In addition, Brenner tumors often have mucinous epithelial cells lining the center of transitional cell nests, and they occasionally develop a discrete mucinous component identical in other aspects to mucinous cystadenomas. A Brenner tumor is a type of adenofibroma in which nests of transitional epithelium grow in fibrous stroma. The coexistence of a mucinous cystadenoma and Brenner tumor supports the theory of a common origin either from coelomic epithelium or remnants of the embryonic mesonephric system.[13] In the present case series, we report a rare combination of a Brenner tumor with serous cystadenoma.

A serous cystadenoma coexisting with a benign Brenner tumor is very rare. This combination suggests a common Mullerian histogenesis.[2] Serous cystadenomas are unilocular or multilocular tumors filled with clear fluid, and the interior and exterior surfaces are usually smooth with focal papillary excrescences on the interior surface. Microscopically, they are lined by flat-to-low columnar ciliated or nonciliated epithelium. Serous tumors can have a fibrous component in the subtype-serous cyst adenofibroma. Histologically, in serous cyst adenofibroma, both epithelial and fibrous components are closely intermixed. However, in our case, there was a clear demarcation between the fibrous solid and serous cystic parts. Cystic changes can occur in fibromas as a degenerative change, in which case, no lining of the epithelium will be seen in the cystic part. In the present case, the cystic part was lined by flattened/cuboidal epithelium all over the walls, including the interface of the solid and cystic parts. The lining epithelium of the cyst was flat to cuboidal rather than ciliated columnar. Although benign serous tumors are typically lined by an epithelium similar to that of the fallopian tube with ciliated and less frequently nonciliated secretory cells, cysts with flattened lining may be seen and represent desquamation of the lining epithelium.[14] A Brenner tumor is composed of nests of transitional epithelial cells surrounded by fibrous stroma, with round-to-oval nuclei with a longitudinal groove and clear-to-eosinophilic cytoplasm.

Proliferating Brenner tumors are characterized by mild-to-moderate nuclear atypia and a complex papillary configuration but without any stromal invasion. A typical Brenner tumor consists of epithelial nests with transitional (urothelial) features occurring in the ovary, usually within a circumscribed nodule of fibrotic stroma. Microscopic transitional cell nests or microscopic Brenner tumors are the immediate precursors of larger Brenner tumors.

The incidence of serous cystadenomas with Brenner tumors, proliferating Brenner tumors, and bilateral Brenner tumors is very rare in literature. To the best of our knowledge, this is only the third reported case with this rare combination with serous cystadenoma.

Conclusion

These cases are important because of the rarity of the combination of a Brenner tumor of the ovary with another ovarian lesion, bilaterality, the proliferative nature, and detection as an incidental finding on histopathological examination. Clinically, because of the presence of solid and cystic component, these tumors may be mistaken as malignancies and radical surgery may be performed. As both solid and cystic components are benign, excision is curative. We report these cases with the aim of creating awareness among pathologists and gynecologists about the occurrence of these rare combinations of ovarian tumors so that misdiagnoses and mismanagement can be avoided.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Scully RE, Clement PB, Young RH, editors. Sternberg's Diagnostic Surgical Pathology. 4th ed. Philadelphia: Lippincott Williams and Wilkins; 2004.
2. Syam Sundar B, Shanthi V, Rao NM, Grandhi B, Reddy VC, Swathi S. Brenner tumor with serous cystadenoma – An unusual combination: A case report. Int J Med Res Health Sci 2015;4:248-50.

3. Green GE, Mortele KJ, Glickman JN, Benson CB. Brenner tumors of the ovary: Sonographic and computed tomographic imaging features. J Ultrasound Med 2006;25:1245-51.
4. Moon WJ, Koh BH, Kim SK, Kim YS, Rhim HC, Cho OK, *et al.* Brenner tumor of the ovary: CT and MR findings. J Comput Assist Tomogr 2000;24:72-6.
5. Borah T, Mahanta RK, Bora BD, Saikia S. Brenner tumor of ovary: An incidental finding. J Midlife Health 2011;2:40-1.
6. Hemalatha AL, Konanahalli P. Bilateral malignant Brenner tumor of ovary. J Obstet Gynecol Indian 2005;55:81-2.
7. Kotsopoulos IC, Xirou PA, Deligiannis DA, Tsapanos VS. Coexistence of three benign and a borderline tumor in the ovaries of a 52-year-old woman. Eur J Gynaecol Oncol 2013;34:186-8.
8. Seidman JD, Khedmati F. Exploring the histogenesis of ovarian mucinous and transitional cell (Brenner) neoplasms and their relationship with Walthard cell nests: A study of 120 tumors. Arch Pathol Lab Med 2008;132:1753-60.
9. Vranes HS, Klaric P, Benkovik LB, Pirkic A. Brenner tumour of the ovary. Acta Clin Croat 2005;44:271-3.
10. Dougherty D, Onyemkpa C, Engel M, Oyasiji T. A case report of an incidental Brenner tumor found after resection of a large ovarian mucinous neoplasm. Int J Surg Case Rep 2018;49:40-3.
11. Fox H, Wells M, editors. Surface epithelial stromal tumor of the ovary. Haines and Taylor Obstetrical and Gynaecological Pathology. Madrid, Spain: Churchill Livingstone; 2003. p. 42.
12. Hiwale KM, Bakane BC. Mixed ovarian neoplasm: Brenner tumor with chocolate cyst. Antiseptic 2002;99:287-8.
13. Sridevi S, Rao VM, Satish Kumar S, Bhagyalakshmi A. Mucinous cystadenoma with Brenner tumor: A case report. J Evid Based Med Healthc 2015;2:455-8.
14. Jayalakshmy PS, Poothiode U, Krishna G, Jayalakshmy PL. Ovarian fibroma with serous cystadenoma-an unusual combination: A case report. Case Rep Obstet Gynecol 2012;2012:641085.