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Case Report

Bilateral incidental benign Brenner's tumors with unilateral mucinous cystadenoma of ovary – a rare case report

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ABSTRACT

Brenner tumors are unilateral and in 5-7% of cases the tumors are bilateral. These tumors are usually identified incidentally during imaging studies or surgery in post-menopausal women. Histologically, BTs are characterized by well-circumscribed nests of transitional epithelium surrounded by a fibromatous background. Brenner tumors are known to coexist with mucinous tumors of ovary. We have presented a rare case in 43-year-old pre-menopausal women presented with heavy bleeding reported as bilateral incidental Brenner's tumor associated with unilateral mucinous cystadenoma along with other pathologies like benign endometrial polyp, adenomyoma and paratubal cyst. It highlights the awareness for extensive sampling of bilateral ovaries because of the common association with mucinous cystadenomas.

Keywords: Brenner tumor, Mucinous cystadenoma, Adenomyoma

INTRODUCTION

Brenner tumors (BTs) are surface-epithelial stromal cell tumors that account for 1–5% of epithelial ovarian tumors.[1] and 1.4-2.5% of all ovarian tumors.[2,3] The majority of these tumours are benign with a 2% incidence of malignancy and occur mostly in the postmenopausal women. The average age of presentation is 50 years with 71% of the patients being more than 40 years.[4] This tumour mostly occurs in the postmenopausal women. Most of Brenner tumours are unilateral and in 5-7% of cases the tumours are bilateral.[5]

These tumors are usually identified incidentally during imaging studies or surgery in post-menopausal women. Histologically, BTs are characterized by well-circumscribed nests of transitional epithelium surrounded by a fibromatous background.[2]

Brenner tumors are known to coexist with mucinous ovarian tumors.[6] Seidman et al observed 1.3-4% incidence of coexisting Brenner tumor and mucinous cystadenomas.[7] A serous tumor is rarely found coexisting with a benign Brenner tumor.[8]

We hereby present a case with concurrence of bilateral Brenner tumor with unilateral benign mucinous cystadenoma of ovary as an incidental finding during Abdominal hysterectomy with bilateral salpingectomy and right oophorectomy in a 43-year-old female done for heavy menstrual bleeding, which showed adenomyoma and benign endometrial polyp on histopathological examination.

CASE REPORT

43-year-old premenopausal women presented in the Gynecology OPD of a private hospital with complaint of heavy menstrual bleeding.

Ultrasound of abdomen and pelvis revealed a bulky uterus with posterior wall fibroid measuring 59×58×52 mm and right ovarian complex cyst was reported containing

multiloculated cyst with solid areas measuring 83×74×58 mm. left ovary was unremarkable. Endometrium thickness was 50 mm. No history of diabetes or hypertension was there. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was planned for this patient as there were large fibroid and right ovarian complex cyst.

Figure 1: Multiloculated right ovarian cyst.

The post-operative specimen received was of right ovarian cyst along with uterus, cervix, bilateral fallopian tubes and left ovary. On gross examination uterus was enlarged weighing approx. 300 gm. Endometrial thickness was 50 mm, and myometrium was trabeculated with single fibroid measuring 2×2cm. cervix had nabothian cyst. right ovary measured 8×6×6 cm. Cut section was multiloculated (Figure 1) with clear fluid and solid mural nodule measuring 4 cm (Figure 2). It was firm to hard in consistency and had yellowish white appearance. No papillary areas detected. Left ovary externally unremarkable measures 3x2x1 cm and cut surface showed normal ovarian tissue along with a well circumscribed tiny solid lesion measuring 1 cm, firm, yellowish white appearance (Figure 3). Right Fallopian tube showed para tubal cyst and left was unremarkable.

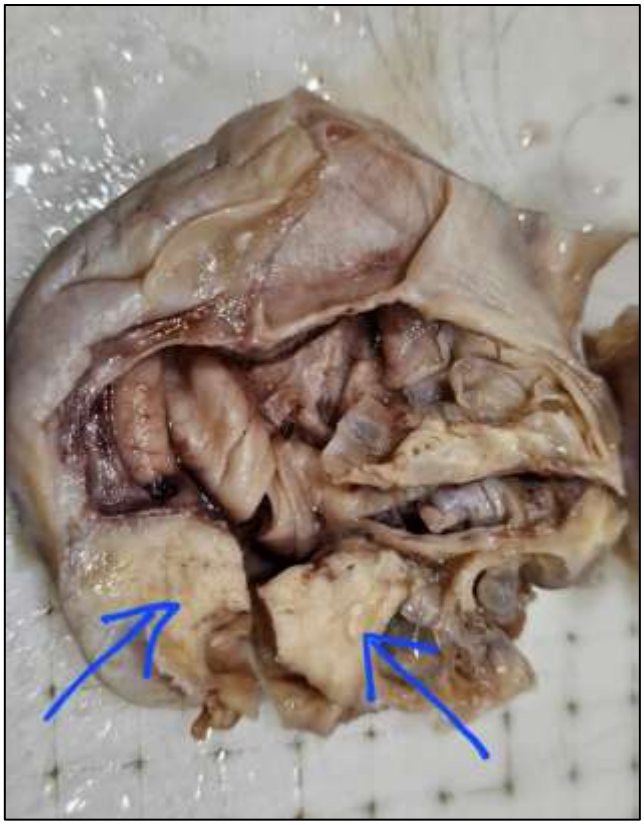

Figure 2: Right ovarian cyst with solid mural nodule (arrow).

Figure 3: Normal ovarian tissue along with a well circumscribed tiny solid yellowish white nodule (arrow).

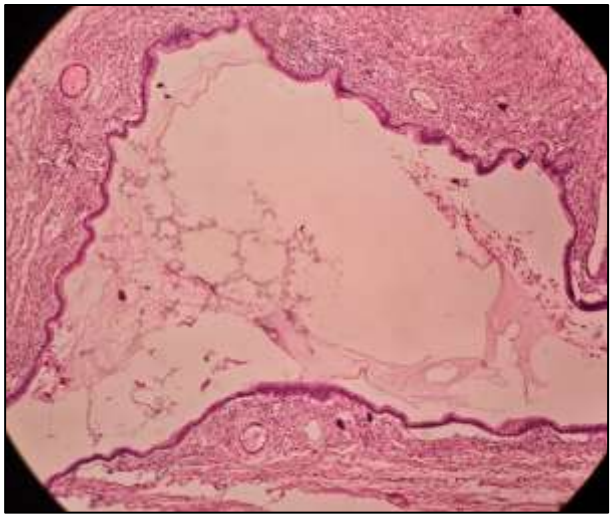

Figure 4: Right ovary showed multiloculated cysts lined by benign mucinous epithelium (100x).

Microscopic examination of right ovarian tumor showed multiple cysts lined by mucinous epithelium (Figure 4). Few solid and cystic nests of urothelium like cells surrounded by dense fibrous stroma were seen (Figure 5). The epithelial cells were well defined, uniform, and polygonal with pale cytoplasm, prominent single nucleolus and grooved nuclei giving a coffee bean appearance (Figure 6). Foci of calcification were present (Figure 7). No evidence of nuclear atypia or mitosis was seen. Sections from left ovary showed tiny nodule of solid and cystic nests of urothelium like cells with fibrous stroma and it was surrounded by unremarkable ovarian stroma (Figure 8). The above microscopic findings were consistent with bilateral benign Brenner tumor associated with benign mucinous cystadenoma in right ovary. Sections from endometrium shows features of benign endometrial polyp (Figure 9). Myometrium shows foci of adenomyosis and adenomyoma (Figure 10). Endocervix showed chronic cervicitis, Nabothian cyst and squamous metaplasia. One of the fallopian tubes showed para tubal cyst.

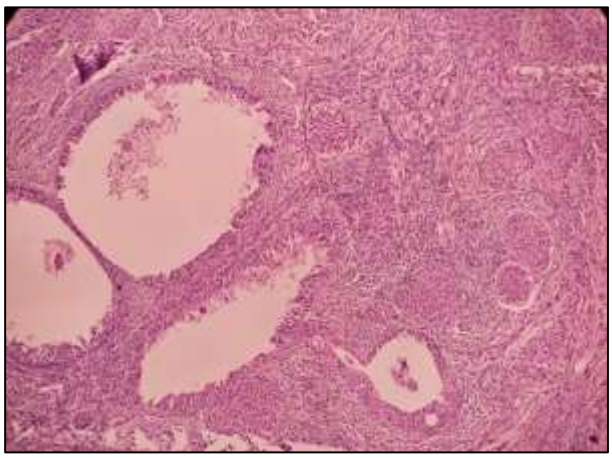

Figure 5: Solid and cystic nests of urothelium like cells surrounded by dense fibrous stroma (100x).

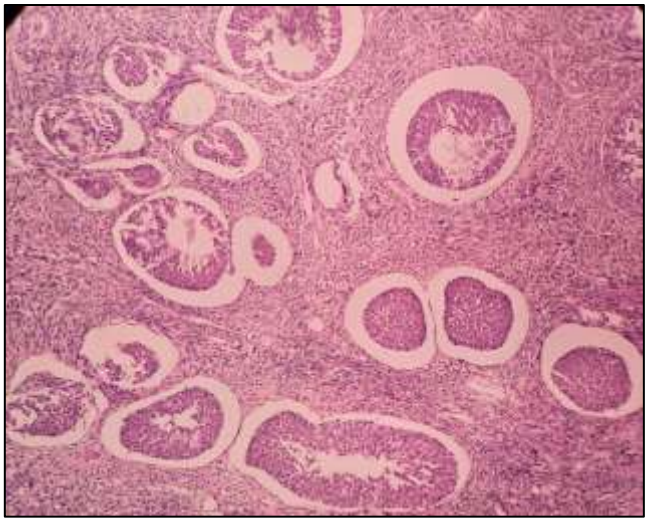

Figure 6: Epithelial cells were well defined, uniform, and polygonal with pale cytoplasm, prominent single nucleolus and grooved nuclei giving a coffee bean appearance (100x).

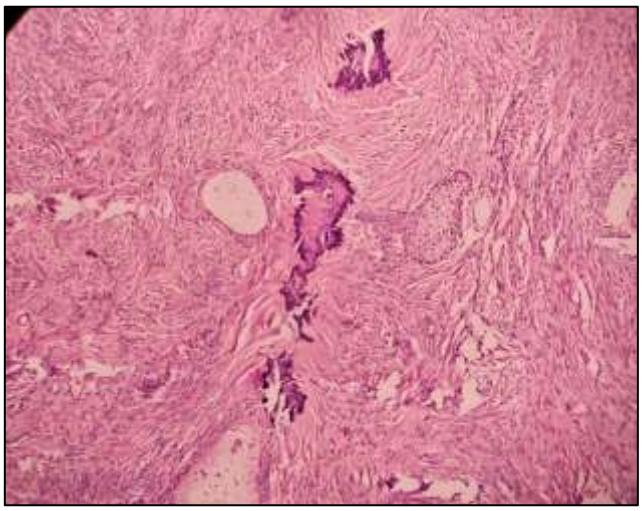

Figure 7: Foci of calcification (400x).

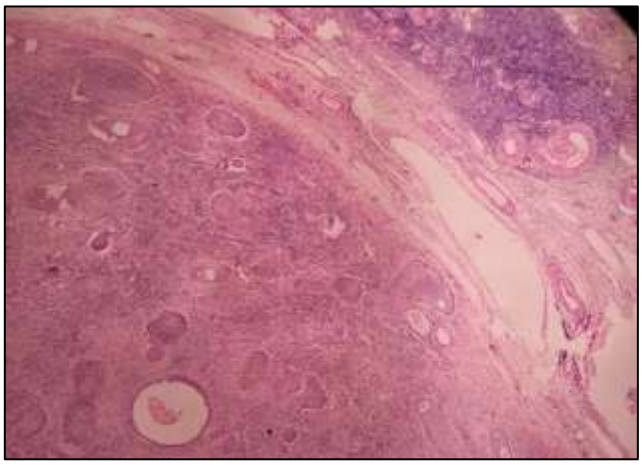

Figure 8: Left ovary with BT and normal surrounding ovary (4x).

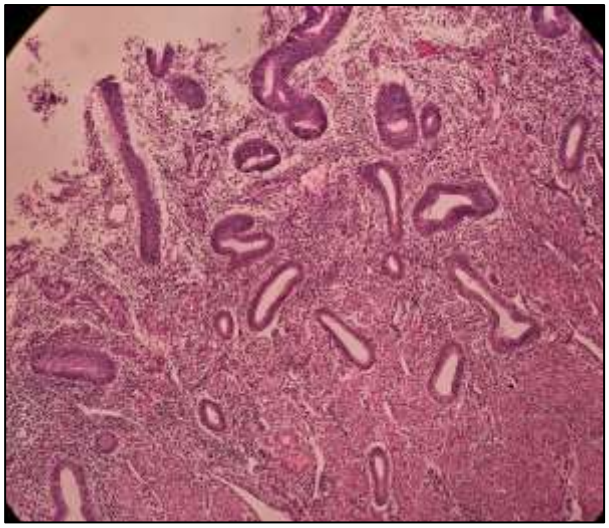

Figure 9: Benign endometrial polyp (100x).

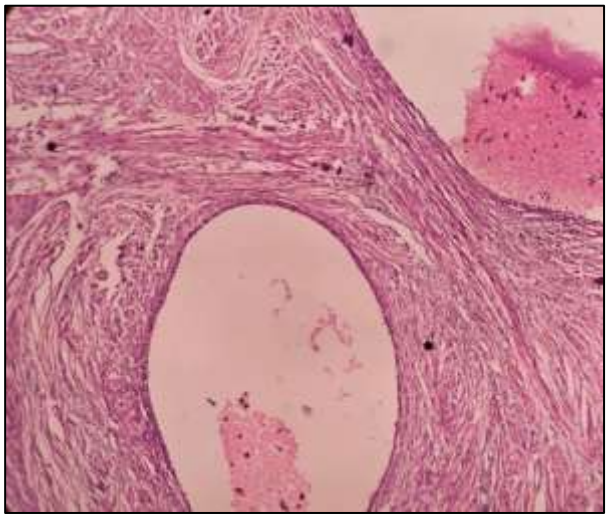

Figure 10: Foci of adenomyosis containing blood and macrophages in lumen (400x).

DISCUSSION

BTs of the ovary make up approximately 5% of all epithelial ovarian neoplasms.[9] Most BTs of the ovary are unilateral. The percentage of bilateral tumors ranges between 3.7% and 8% in various case series. 11% in a study by Alloush et al.[10] They are typically multilocular or unilocular solid and cystic tumors with variable solid nodules and papillary excrescences in the cystic parts.[1] On gross examination Brenner tumors are solid, well circumscribed, pale yellow to grey white in color with a solid or fibromatous cut surface. The tumors vary in size from 1 cm to 30 cm.[11]

We have presented a case of bilateral incidental benign Brenner tumor out of which one ovary was enlarged, cystic multiloculated and Brenner tumor was seen as mural solid nodule (4 cm) (Figure 2) whereas the other ovary was of normal size which shows tiny nodule in ovarian parenchyma (1cm) (Figure 3).

BTs have a nonspecific appearance on USG and Computed Tomography. Benign BTs are asymptomatic and are typically found incidentally during imaging studies and surgeries performed for other purposes.[2] Among symptomatic patient's common symptoms include vaginal bleeding, a palpable pelvic mass and pelvic pain.[12] In our case patient presented with heavy bleeding and she was diagnosed on USG with fibroid and right ovarian complex cyst. On microscopy, apart from incidental BT and mucinous cystadenoma she also had adenomyosis, adenomyoma, benign endometrial polyp and Para tubal cyst which is rare association.

Benign BTs are composed of nests of transitional epithelium with longitudinal grooves in a fibromatous background. Central cystic changes with mucinous metaplasia of the luminal aspect are common findings. Hence, it was postulated that overgrowth of the mucinous component accounts for the common association between BTs and mucinous cystadenomas.[13]

Our case on microscopy showed islands of transitional epithelial cells with nuclear grooves lying in fibromatous stroma. Features of malignany were not seen. Most BTs occur unilaterally and scattered punctate calcifications are a common finding on CT and sonographic scans.[14] Our case had many scattered foci of calcification among fibromatous stroma. Alloush et al six of nine cases of BT had a normal intra-operative appearance of the involved ovary which suggests that the normal intra-operative appearance of the contralateral ovary in mucinous cystadenoma cases does not exclude the possibility of BT and bilateral oophorectomy might be considered in appropriate scenarios.[10]

In our case one BT was like mural nodule with mucinous cystadenoma whereas other BT was like tiny nodule in normal sized ovary. BTs are associated with endometrial hyperplasia in 4-14% of cases due to testosterone and estrogen release.[15] In our case USG reported endometrial hyperplasia of 50 mm and on microscopy it was benign endometrial polyp.

CONCLUSION

Most ovarian BTs are commonly detected as incidental tumors in surgical Gynecology specimens done for other commoner lesions and they are Benign in nature. Pathologists and surgeons need to be aware of the common association between mucinous cystadenomas and BT as small nodules of BT can be easily overlooked due to the large size and cystic consistency of mucinous cystadenomas in most cases. This case report highlights the occurrence of incidental nature of BT which can occur as mural nodule in enlarged cystic ovary as well as in normal sized ovary creating awareness for extensive grossing for these lesions.

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