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Case Report

Borderline Brenner Tumour: A Rarely Encountered Clinico-Pathological Ovarian Entity: Case Report

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Abstract

Borderline Brenner Tumour of the ovary is a very rare neoplasm, seldom encountered in clinical practice, as the incidence of Brenner Tumours itself is very low. They are generally small and incidentally detected solid tumours. However, we report the case of a large solid-cystic tumour in a symptomatic post-menopausal lady with normal tumour markers. The clinical and radiological picture intersected with features of both benign and malignant ovarian tumour. She underwent staging laparotomy and complete surgery. Intra-operative frozen evaluation favoured a benign mass. The diagnosis of Borderline Brenner Tumour was clinched with a combined evaluation of histomorphology and immunohistochemistry (IHC), thus outlining the role of careful evaluation to diagnose this rare entity, to optimise post-operative management.

Keywords: *Borderline Ovarian; Brenner Tumour; Low-Grade Brenner; Transitional Cell Tumour; Borderline Brenner; IOTA ADNEX*

Introduction

Brenner Tumour of the ovary was first described over a century ago, as a surface epithelial neoplasm. It is composed of urothelial type lining, accounting for only 1.5 - 2% of the ovarian neoplasms. It is classified into benign, borderline, and malignant by WHO [1]. While majority (90 - 95%) are benign, a borderline, and malignant histology is seen in 3 - 4% and 1% respectively [2]. Although asymptomatic and incidentally detected at pathology, they may present with a large abdomino-pelvic mass and/or vaginal bleeding. Our case was a post-menopausal lady in whom the diagnosis of borderline Brenner tumour (BBT) was confirmed on histopathology and IHC.

Case Report

A 61 y.o. parous lady presented with post-menopausal bleeding of 3 - 4 months duration and abdominal heaviness. There was no significant medical or family history. Examination revealed a tense cystic mass up to umbilicus, more on the right side with no free fluid in abdomen. The same findings were confirmed on vaginal-rectal examination, with a normal sized uterus felt posterior to the mass. Patients are seen to delay care seeking during the COVID pandemic, as was in this case. Outpatient Pipelle biopsy was done at presentation and other work up advised. Tumour markers were CA-125 10.4U/mL, CEA < 0.5 ng/mL, AFP 7.05 ng/mL, LDH 241 U/L and beta-HCG 1.2 mIU/mL. FSH and LH were in menopausal range (35 mIU/mL and 16 mIU/mL respectively) but S. E2 was high (73.4 pg/mL). Ultrasound revealed a well-defined large complex cystic lesion 15.9 x 10.8 x 15.5 cm size with multiple echogenic nodular solid areas along posterior wall, largest area measuring 5.9 x 4.6 cm, suggestive of malignant ovarian mass arising from right ovary (Figure 1). There was no ascites or lymphadenopathy. PET-CT was done and showed a well circumscribed thin walled solid-cystic (predominantly cystic) lesion 16.1 x 13.9 x 11.2 cm, with the multiple solid components showing contrast enhancement and FDG avidity (Figure 2). There was no other FDG avid

disease elsewhere in the body. She was planned for surgery; however, she did not want to get it done due to the pandemic. She presented again after 2 months, repeat ultrasound showed no significant interval changes in the mass (size 16.6 x 15.4 x 11.4 cm, volume 1540cc). Entering the relevant variables into the IOTA ADNEX model [3] showed an 83.1% risk of malignancy, with the highest relative risk for borderline tumour. Outpatient Pipelle showed proliferative endometrium. Time from initial consultation to surgery was 64 days. She was consented for staging laparotomy, total hysterectomy, bilateral salpingo-oophorectomy, infra-colic omentectomy and frozen evaluation. Intra-operatively, there was no ascites, peritoneal washings were collected. Systemic inspection and palpation of abdomino-pelvic cavity did not reveal any tumour deposits or lymphadenopathy. The 20 x 17cm solid-cystic right ovarian mass was removed intact followed by definitive surgery. Frozen section from a papillary area in the mass was reported as favouring benign. Peritoneal fluid cytology was negative for malignant cells. She made uneventful post-operative recovery. Final histopathology showed low grade papillary urothelial neoplasm, crowded nests of uniform transitional epithelium, occasional mitosis with no stromal invasion. Cyst wall was lined by bland uniform cuboidal epithelium. Endometrium was hyperplastic with no atypia and omentum was free of tumour. IHC showed strong CK positivity (4+), CK7+/CK20-, diffuse nucleolar expression of p63 in tumour cells, and negative for Uroplakin, WT-1 and GATA-3. Ki67 expression was positive in 30-40% of neoplastic cells. This indicated of atypical proliferative Brenner tumor with intraepithelial carcinoma. Diagnosis of Borderline Brenner Tumour Stage IA was confirmed. No adjuvant treatment was recommended. She is doing well 8 months after surgery with no recurrence.

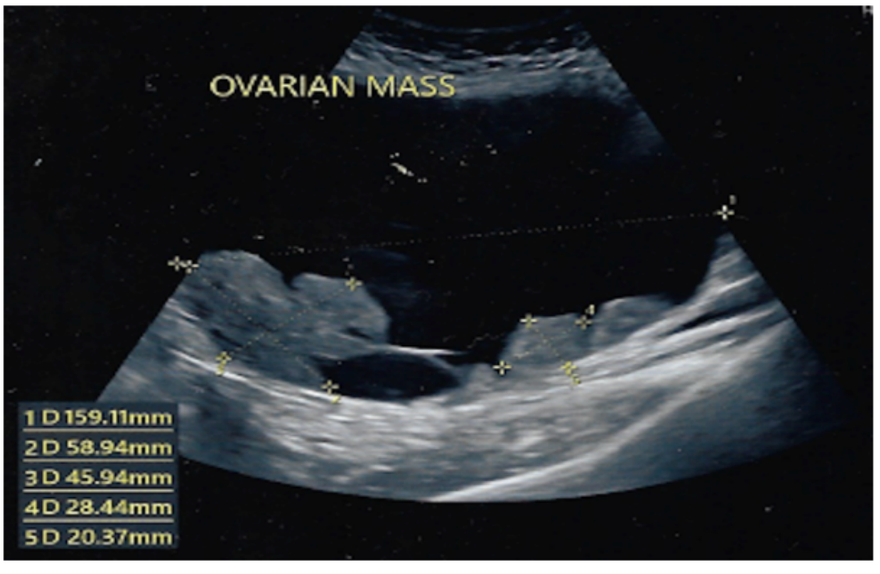

Figure 1: *Ultrasound showing well-defined large complex cystic lesion with multiple echogenic nodular solid areas along posterior wall.*

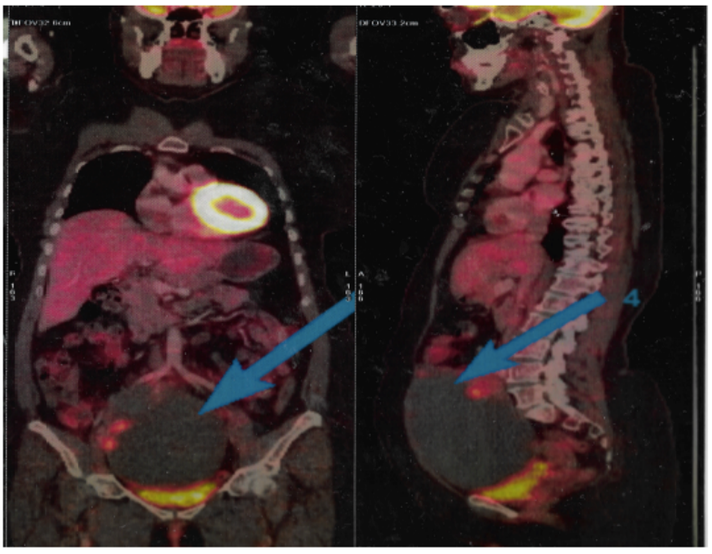

Figure 2: *PET-CT showing a well circumscribed thin walled solid-cystic lesion showing contrast enhancement and FDG avidity.*

Discussion

Transitional cell tumours of the ovary were first described by Fritz Brenner in 1907. Borderline Brenner tumours (BBT) were first described in 1971 as proliferating Brenner tumours by Roth and Sternberg. Since then, less than 60 cases have been described in literature as per Zheng., *et al.* who also describe BBT as having epithelial proliferation more than seen in benign Brenner's tumour, but without stromal invasion [4]. There is no specific tumour marker for it unlike other epithelial tumours of the ovary.

In a multicentric retrospective case review, 30 cases of BBT were reviewed [5]. The mean age of presentation was 69 years (range 52 - 84 years). Unilaterality and Stage I disease was the norm, with no stromal microinvasion. Only 1 case of recurrence was described in this data set.

Although benign Brenner tumours are similar to other solid tumours of the ovary on imaging, borderline and malignant forms are usually cystic with papillomatous projections/ mural nodules in the cavity [6], as in this case. On histopathology, fibrous stroma is more common in benign tumours, and less conspicuous in borderline and malignant forms, limited to the solid projections in the mass [7]. These papillary projections are covered with transitional epithelium, with some mitosis, but no necrosis or stromal invasion. The thecoma/fibroma component may secrete estrogen, as in this case, and lead to symptoms of endometrial hyperplasia and post-menopausal bleeding. Immunohistochemistry plays a role in distinguishing borderline and malignant forms from transitional cell carcinoma of ovary.

The staging for BBT is the same as for malignant ovarian tumours. Peritoneal staging and lymphadenectomy are not recommended, as all reported patients (but one) have presented with stage 1 disease. Surgical resection of the tumour is curative as they are organ confined [8].

Conclusion

In summary, this patient had features of malignant disease - short history, FDG uptake on PET and large solid cystic ovarian mass, as well as benign disease - normal tumour markers, no ascites, peritoneal deposits or lymphadenopathy. Even a delay of two months due to the pandemic did not upstage the disease, thus highlighting that these rare ovarian tumours need appropriate characterisation based on a combination of clinical, radiological and pathological factors, to guide optimum management.

Informed Consent

Consent to publish was taken from the patient in prescribed format.

Disclosures

None.

Conflict of Interest

None.

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