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Case Report

Benign Brenner's tumor of ovary: a case report with review of literature

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ABSTRACT

Brenner's tumor of the ovary is a rare neoplasia with an incidence of 02-03% of all ovarian tumors. Its origin is controversial; the new hypothesis describes it as transitional cell metaplasia of Walthard cell nests embedded in the ovary. The world health organization has classified it into benign, borderline, and malignant varieties, with incidences of 95%, <5%, and <2%, respectively. Benign Brenner's tumor (BBT) is usually <2 cm in size, unilateral, solid, grayish white, well-circumscribed, rubbery to firm nodule, arising from ovarian cortex and resembling fibroma or thecoma of ovary. The average age of patients is 50 years. Diagnosis of the pathology is challenging as it does not have a specific clinical presentation or investigatory findings, and is usually diagnosed incidentally on histological examination of the ovary resected with other genital organs. Microscopy of the tumor shows a fibro-epithelial solid mass consisting of transitional epithelial cell nests surrounded by dense fibrous tissue. No further treatment is required in benign cases, whereas chemotherapy with or without radiotherapy and follow-up is needed in borderline and malignant cases. A case of unilateral BBT of the ovary in a 42-year-old parous lady was diagnosed in this institution, who had undergone total abdominal hysterectomy with bilateral salpingo-oophorectomy for fibroid uterus with abnormal uterine bleeding (AUB) and severe anemia. The presentation aims to report this rare pathology of Brenner's tumor without a specific clinical presentation, diagnosed incidentally on histopathological examination (HPE), and to add to the statistics.

Keywords: Ovarian tumor, Transitional cell metaplasia, Fibroma, Thecoma

INTRODUCTION

Benign ovarian tumors are common neoplasia in women and account for about 75 - 80% of total ovarian tumors in the reproductive age group.[1] They are classified histologically and vary in incidence, and their presentations are variable due to size or hormone production. Brenner's tumor was first described by a German surgeon and pathologist, Fritz Brenner, in 1907, which is named after him.[2] WHO has classified it into three groups as benign, borderline, and malignant on histological features.[3] BBT is the commonest among the three types.[4] Malignancy is seen in <02% cases, whereas borderline tumors are detected in <05% of total Brenner's tumors.[5] It is a rare neoplasia of the ovary and constitutes 2-3% of all ovarian tumors.[6] The tumor also has an extra-ovarian origin. Though it has a predilection for postmenopausal women, with an average age incidence of over 50 years, it can affect younger women.[7] The tumor is usually <2 cm in size, solid, grayish white, well-circumscribed, rubbery to firm in consistency, arising from ovarian cortex, resembling fibroma or thecoma of ovary. In 10% of cases, tumors of size >10 cm may be detected, which are partially cystic.[8] Bilateral BBT may be found in 5%-7% of cases.[9] Microscopically, it is a fibro-epithelial solid tumor consisting of transitional epithelial cell nests surrounded by dense fibrous tissue. Large tumors may contain cystic spaces. Clinical presentation is variable, from asymptomatic to pelvic discomfort, depending on size and presence of other histological components.

Diagnosis of ovarian Brenner's tumors at present is challenging, as it does not show typical clinical, laboratory, or radiological features. The tumor is diagnosed incidentally on HPE of the ovary resected with other pelvic organs for some indications. With this background, we present a case of BBT of the ovary diagnosed on HPE of resected specimen in a total abdominal hysterectomy with bilateral salpingo-oophorectomy for a fibroid uterus with AUB. It highlights the importance of HPE of the resected organ for a definitive diagnosis and subsequent management. The aim of the presentation of this case is to report the rarity of the tumor, the absence of specific clinical, biochemical, or imaging features, and to add to statistical accounting.

CASE REPORT

A 42-year-old lady reported to the gynecology outpatient department of the institution with the complaint of heavy menstrual bleeding for one and a half years. There was no history of passing clots per vagina, foul-smelling vaginal discharge, dyspareunia, fever, or bowel and bladder problems. Her present menstrual cycle was 10 days of heavy bleeding, followed by spotting for the rest of the month till she got her next menstrual bleeding. She was treated by a local healthcare facility with oral medication. However, she did not get relief, for which she was referred to this institution. The patient is an illiterate lady from a rural area and belongs to a low socioeconomic status. Her past menstrual history was 4/30 days, regular, normal flow, and no pain. She got married at the age of 13 years, para two with both caesarean deliveries and concurrent tubal sterilization during the second caesarean section at the age of 28 years. There was no past or family history relevant to her present sickness. She did not have any history of an allergy, addiction, or blood transfusion. On arrival at the hospital, she was found to be physically and mentally stable. There was severe pallor; no icterus, edema, or lymphadenopathy. Her pulse was 80/min, BP-120/80 mm Hg, SpO_2 98% in room air, afebrile, respiration 16/min. Abdominal examination revealed a healthy sub-umbilical midline scar. The abdomen was soft and nontender. There was no distension or mass lesion. Clinically, there was no abnormality in the cardiovascular, respiratory, and central nervous systems. Per speculum examination revealed minimal bleeding from the uterus; a fleshy mass of about 4x5cm protruding at the external os, arising from the uterine cavity with a pedicle which did not bleed on touch; nor was there any infective or foul-smelling discharge. Bimanual examination (BME) demonstrated a bulky cervix. The uterus was bulky, nontender, and mobile, in mid position, and the fornices were free. Both a Papanicolaou (PAP) smear before BME and a biopsy after BME from the mass at the external os were taken. On investigation, her hemoglobin was 05. 0gm% with a low hematocrit, and there were features of microcytic and hypochromic anemia in the peripheral smear. White cell and differential count did not show features of infection. Fasting and postprandial blood sugar were 105 and 500 mg/dl, respectively, and negative for urinary ketone bodies. Renal function, liver function, serum lipids and electrolytes, and coagulation profile were within normal limits. She had nonreactive serum HIV and HBsAg. Her blood group was B positive. X-ray chest PA view, ECG, echocardiography, and urine test report were within normal range. Fundoscopy was within normal limits. Transabdominal and transvaginal ultrasonography (USG) reported a bulky anteverted uterus, endometrial thickness of 4mm, and a bulky cervix with a 4.5×2.5 cm heterogeneous mass in the canal, and no adnexal lesion. The PAP smear did not show an intraepithelial lesion or malignancy, and the biopsy report indicated leiomyoma without evidence of malignancy. Under the federation of obstetrics and gynecology (FIGO) classification of AUB, she was diagnosed as AUB-L(leiomyomas), P (polyp), with severe anemia and diabetes mellitus. She was treated with two units of packed cells, one unit of whole blood transfusion at intervals, and oral hypoglycemic drugs. She was planned for a Total abdominal hysterectomy with bilateral salpingectomy, keeping her informed about other pathology and extension of surgery if required. With fitness and informed consent, laparotomy through the previous abdominal scar under regional anesthesia was done. Laparotomy revealed a uterus of 11×6 cm, mild enlargement of the right ovary with a firm to hard nodule. Both the fallopian tubes and the left ovary appeared healthy. Fibroid polypectomy was done through an incision in the uterus before clamping the uterine artery pedicle. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was done considering the suspicious right ovary. On examination of the specimen, the size of the right ovary was 4.0×3×2 cm, containing a mass of 2.5×02 cm, which was solid, rubbery to firm in consistency, grayish-white in color, as shown in Figure 1. Cut section of this rubbery mass showed a grayish solid mass without any cystic area, as reflected in Figure 2. Her intra- and postoperative periods were uneventful. Histopathology of the specimen reported a BBT in the right ovary, as presented in Figure 3, papillary cervicitis, leiomyomas, and endometrium in the proliferative phase. The patient was healthy and doing well on the subsequent follow-up visit. She has been advised to consult a physician for the control of her blood sugar.

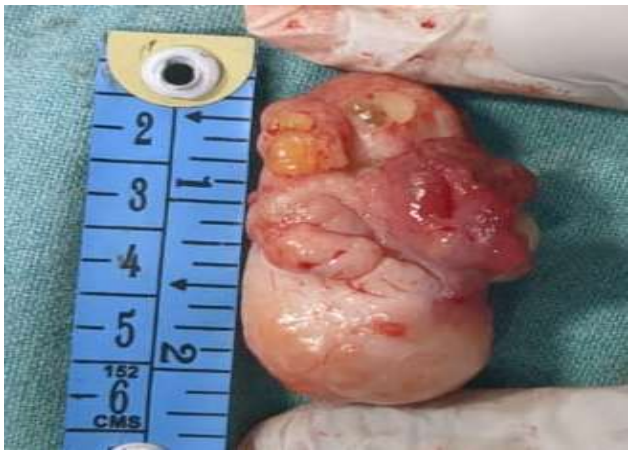

Figure 1: Right ovary including the solid mass.

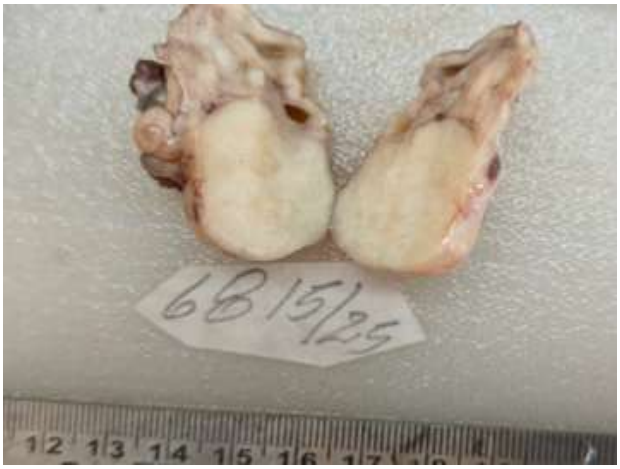

Figure 2: Cut section of the right ovary including the mass.

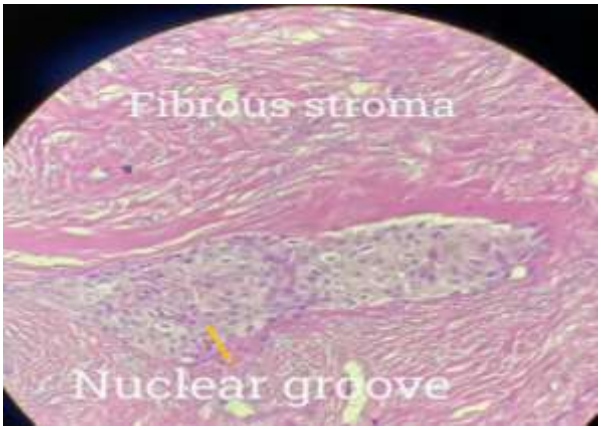

Figure 3: Histopathological picture of the tumor showing a nest of transitional epithelium with nuclear groove and surrounding fibrous stroma.

DISCUSSION

Incidence of BBT is 95% of all Brenner's tumors.[10] It is a rare solid ovarian tumor that may be associated with serous or mucinous cystadenoma in 30% of cases.[5] Origin of Brenner's tumor is controversial. As per the old hypothesis, it arises from ovarian surface epithelium by transitional cell metaplasia, whereas the new hypothesis considers that the origin is from transitional cell metaplasia of Walthard cell nest, which are cells at the tuboperitoneal junction that are embedded in the ovary.[11] Though the usual age incidence is the fifth to seventh decade of life, it may develop <30 to >80 years of age.[12,13] Our patient was 42 years old. The size of BBT of the ovary is usually <10 cm, and in our case, it was 2.5×2.0 cm.[8] It is usually an incidental histopathological finding without any associated symptoms, though the patient may present with vaginal bleeding, pelvic pain with or without an adnexal mass.[14] Our patient presented with AUB, but as the mass was not large enough to produce pelvic congestion, nor was the tumor hormone-producing, the presence of other pathologies like submucous leiomyoma and cervicitis might be responsible for her symptoms. Resemblance of the stromal component of the tumor to ovarian theca cells is reported to produce estrogen from aromatization of androgen, which may cause these hormone-related problems in the patient.[13] No definite tumor marker has been associated yet with BBT, though authors have reported elevated CA-125 in malignant Brenner's tumor, which reduced after treatment.[15] Preoperatively, we did not do serum CA-125 as no adnexal mass was reported clinically or in USG. Neither ultrasonography nor computerized tomography is specific in diagnosing this tumor, as a solid adnexal mass is reported by these imaging methods, with differential diagnosis of fibroma, fibro-thecoma, or pedunculated fibroid in these cases.[15,16]

CONCLUSION

BBT is usually a small, unilateral, and solid tumor that is without a definite clinical presentation or diagnostic investigations. It should not be disposed of without HPE. It may be in borderline or malignant form, which needs further management with a good prognosis. Histopathology is the gold standard for diagnosing the pathology as well as for management. The immunohistochemical test is valuable in cases with borderline and malignant characteristics.

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