A case report of an incidental Brenner tumor found after resection of a large ovarian mucinous neoplasm

Danielle Dougherty a,*, Chibueze Onyemkpa a, Michael Engel b, Tolutope Oyasiji c

a Michigan State University, Department of Surgery, 1200 East Michigan Ave., Suite 655, Lansing, MI 48912, United States
b McLaren Medical Group, 2487 North Elms Road, Flushing, MI 48433, United States
c Department of Surgical Oncology, Barbara Ann Karmanos Cancer Institute at McLaren Flint, Wayne State University, 4100 Beecher Road, Flint, MI 48532, United States, & Department of Surgery, Michigan State University

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A B S T R A C T

INTRODUCTION: Brenner Tumors are rare adenofibromas that are most commonly benign and discovered in post-menopausal women.

PRESENTATION OF CASE: This is a case report of a 57-year-old female with three months of progressively worsening abdominal pain due to a large abdominal mass discovered on CT scan. Surgical removal of the mass revealed a giant mucinous tumor of the ovary with an associated Brenner tumor that was discovered incidentally.

DISCUSSION: Although the Brenner tumor was accurately identified in the intraoperative frozen section evaluation, the mucinous tumor was underdiagnosed by frozen section as benign when permanent section revealed borderline mucinous cystadenoma. This finding did not change the treatment course for this particular patient as she had expressed personal preference for total abdominal hysterectomy. However, underdiagnosis of frozen sections of ovarian tumors is not rare. It is unclear whether an associated Brenner tumor increases malignancy potential.

CONCLUSION: Further investigation is required to determine whether associated Brenner tumors found during frozen section are more highly associated with malignancy and could therefore change intraoperative and overall decision making.

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1. Introduction

Brenner tumors are rare adenofibromas. Their size varies from less than 1 cm to up to 30 cm. They are usually unilateral and are characterized by their epithelial lining that contains clusters of transitional cells that resemble the urinary tract epithelium. Sometimes these nests have microcysts or mucinous glands in the center [1]. Transitional cell tumors of the ovary were described for the first time by Brenner in 1907. They are rare, accounting for only 2% of all ovarian tumors [2]. They are most commonly benign and discovered in post-menopausal women [3]. This is a case report of a Brenner tumor that was discovered incidentally associated with a giant mucinous tumor of the ovary. This work has been reported in line with the SCARE criteria [4].

2. Case presentation

The patient is a 57-year-old female who presented to clinic with a chief complaint of three months of progressively worsening abdominal discomfort. She denied any weight loss or change in eating or bowel habits. As a result of the gross distention of the abdomen and concern for probable gastrointestinal origin of abdominal pathology, a CT scan of the abdomen and pelvis was performed. This demonstrated a well circumscribed mass that extended from deep within the pelvis in the presacral space and extended towards the diaphragm in the left upper quadrant (Fig. 1). The mass measured 40 × 22 × 27 cm in size. It exhibited diffuse septations and had irregular and nodular enhancements. At this time the differential diagnosis included mucinous cyst adenoma or cyst adenocarcinoma of the ovary. CEA and CA-125 levels were elevated at 6.1 ng/mL and 249.4 U/mL respectively. Due to the size of the lesion, it was determined that neither laparoscopic biopsy nor CT guided biopsy was feasible, leaving the option of exploratory laparotomy with intraoperative frozen section to determine the nature of the tumor and the extent of surgical treatment. If the frozen section proved to be malignant, the plan was to perform cytoreductive surgery, including total abdominal hysterectomy, omentectomy, appendectomy, possible intestinal resection, and retroperitoneal lymphadenectomy for completion of staging. The patient also clarified during preoperative discussions that due to personal preference she wished for a total abdominal hysterectomy with bilateral salpingo-oophorectomy whether the frozen sections were benign or malignant.
During the procedure, the tumor was assessed once the peritoneal cavity was accessed. The massive tumor was carefully dissected free from surrounding structures. It was traced to the left adnexa and was closely related to the atrophic left fallopian tube. It was removed and sent for intraoperative frozen section (Fig. 2). Pathological evaluation identified the frozen section as benign mucinous cystadenoma of the left ovary, thereby terminating the procedure. Final surgical pathology of permanent sections reported ovarian mucinous borderline tumor which appeared to be of mixed intestinal and endocervical type with no areas of invasion. The benign pathology is demonstrated in Fig. 3, which shows a combination of intestinal and endocervical cells. Borderline classification of the tumor was characterized by complex mucinous epithelial architecture which was more evident on the permanent sections than on the original frozen section slides which showed no features of borderline tumor (Fig. 4). The focal Brenner tumor cell nests were noted on both frozen and permanent sections.

3. Discussion

Although Brenner tumors are rare, it is not uncommon to find a Brenner tumor associated with a mucinous tumor. The association between the two is reported to be between 1–16% [5]. Although the Brenner tumor is classified by its transitional cells, they often...
contain a central cavity which contains mucinous cell epithelium. It has been proposed that in some instances the mucinous cells proliferate and result in a mucinous tumor which, as it grows larger, has the capacity to compress and obliterate the original Brenner tumor. It has been demonstrated that in combined mucinous and Brenner tumors, the two cell types share a common clonal origin, and that therefore the mucinous tumor may have in fact arisen from the Brenner tumor [6].

It is difficult to use imaging alone to determine if a mass is a Brenner tumor. One retrospective study failed to identify any ultrasound features specific to Brenner tumors, which had been discovered incidentally after adnexal masses of unknown etiology were removed [7]. One identified CT and MRI finding is extensive calcification within a solid component of a cystic mass [8].

In the case report presented, the intraoperative frozen section of the mass appeared to be a benign mucinous tumor with incidental finding of associated Brenner tumor. However, final permanent section evaluation demonstrated a borderline mucinous cystadenoma again associated with focal Brenner tumor cell nests. This frozen section underdiagnosis is not uncommon and one investigation demonstrated that approximately 30.6% of ovarian tumors that were confirmed by permanent section pathological evaluation to be borderline were actually underdiagnosed as benign tumors during frozen section evaluation intraoperatively [9]. However, the use of frozen section for the evaluation of ovarian neoplasms is as a whole very accurate and important for determination of surgical treatment plans intraoperatively. In another study, the overall accuracy of intraoperative frozen section for the evaluation of ovarian neoplasms was found to be above 99% [10].

In this patient’s case, neither the intraoperative frozen section incidental finding of an associated Brenner tumor, nor the post-operative final pathological report of borderline mucinous tumor, changed surgical treatment. This is due to the fact that the patient was post menopausal and desired hysterectomy with bilateral salpingo-oophorectomy no matter what the pathological findings demonstrated, and this was clearly communicated preoperatively. However, in cases of women who wish for a more conservative surgical approach, conservative treatment of borderline tumors of the ovary has been accepted as having good safety and efficacy. Conservative treatment consists of preserving the uterus and at least part of one ovary, combined with comprehensive surgical staging [11]. Despite the importance of attempting to preserve fertility in premenopausal women with this condition, close follow up is essential. One study demonstrated that 13% of mucinous borderline tumors of the ovary recur as invasive carcinoma in 10 years, thereby arguing that a salpingo oophorectomy is preferred over cystectomy alone [12].

It is unclear whether the incidental finding of Brenner tumor upon evaluation of a removed ovarian mass is associated with increased malignancy potential. In this case, the pathological findings of both frozen and permanent sections did not affect patient care or treatment decisions. However, given the combined incidental Brenner tumor finding and underdiagnosed tumor identification of frozen section, it may be beneficial to further explore whether the identification of Brenner tumors have significant implications for the categorization of associated ovarian masses.

**Ethical approval**

This case report is exempt from ethical approval in our institution.

**Consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Author contribution**

Danielle Dougherty, MD: study concept and design, data interpretation, manuscript writing.

Chibueze Onyemkpka, MD: study concept and design.

Michael Engel DO: data interpretation.

Tolutope Oyasiji MD: study concept and design, data interpretation, manuscript writing.

**Registration of research studies**

This does not apply as it is a case report of a patient who has given written consent and has been de-identified. It is therefore not prospective research involving human participants.

**Guarantor**

Dr. Tolutope Oyasiji, MD.

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