

Recurrence of Brenner ovary borderline tumor in the abdominal wall postoperative scar – a case report and research of the literature

Wznowa guza Brennera o granicznej złośliwości w bliźnie pooperacyjnej powłok brzusznych – opis przypadku i przegląd piśmiennictwa

Lukasz Klasa¹, Dariusz Wydra¹, Wojciech Biernat²

¹ Department of Gynecology, Gynecological Oncology and Gynecological Endocrinology, Medical University of Gdansk, Poland

² Department of Pathomorphology, Medical University of Gdansk, Poland

Abstract

We report a case of a 74-year-old female, who underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy due to a solid-cystic tumor of the right ovary.

The histopathological diagnosis revealed a borderline Brenner tumor (BBT). After 25 disease-free months the patient was admitted to a hospital because of a tumor in the postoperative scar of the abdominal wall, later diagnosed as infiltrating Brenner tumor consistent with ovarian borderline lesion. The tumor in the postoperative scar was therefore diagnosed as BBT.

The article presents results of literature search on BBT in order to find characteristic features of this very rare ovarian tumor. To the best of our knowledge, this is the first report of subcutaneous recurrence of Brenner ovary tumor of low malignant potential.

Key words: **Brenner tumor / borderline malignancy / ovary / recurrence / scar /**

Correspondence:

Lukasz Klasa

Department of Gynecology, Gynecological Oncology and Gynecological Endocrinology,
Medical University of Gdansk

ul. Kliniczna 1a, 80-402 Gdansk, Poland

tel.: +48 514 289 398, fax: 0048 58 341 80 03

e-mail: klasik@wp.pl

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Lukasz Klasa et al. *Recurrence of Brenner ovary borderline tumor in the abdominal wall postoperative scar...*

Streszczenie

Artykuł ten opisuje przypadek 74-letniej pacjentki, u której w lito-torbielowatym guzie jajnika prawego po przeprowadzonej operacji przezbrzusznego usunięcia macicy z przydatkami rozpoznano łagodnego guza Brennera. Po 25 miesiącach od pierwotnej operacji chorą przyjęto do szpitala z powodu guza w bliźnie pooperacyjnej w powłokach brzusznych.

W badaniu histopatologicznym zmiany rozpoznano utkanie guza Brennera o granicznej złośliwości (BBT- Borderline Brenner Tumor). Ponowna weryfikacja obrazu histopatologicznego pierwotnego guza jajnika ujawniła wśród dominującego utkania łagodnego guza Brennera obecność ognisk o wzmożonej proliferacji nabłonka typu urotelialnego. Wobec powyższego, diagnozowanego guza powłok uznano za ognisko wznowy uprzednio rozpoznanego guza jajnika. Według naszej wiedzy jest to pierwszy w piśmiennictwie przypadek wznowy skórnej guza Brennera jajnika o granicznej złośliwości.

W artykule znajduje się przegląd literatury dotyczącej BBT. Na podstawie piśmiennictwa autorzy artykułu podjęli próbę znalezienia charakterystycznych cech guzów Brennera w badaniu klinicznym oraz badaniach obrazowych, a także próbę ustalenia rokowania u chorych z tym jakże rzadkim rozpoznaniem.

Key words: **guz Brennera / graniczna złośliwość / wznowa / blizna /**

Introduction

Brenner tumor (BT) of the ovary was originally described by a German pathologist Fritz Brenner in 1906. This uncommon epithelial-stromal neoplasm represents only 1.4-3% of all ovarian tumors [1]. Clinically, it is usually small, solid, unilateral and asymptomatic. BTs are diagnosed accidentally, usually after ovarian resection due to other causes. Infrequently, this neoplasm may manifest by Meigs syndrome, which includes a benign ovarian tumor, accompanied by ascites, hydrothorax, and less often hydropericardium. According to the literature, Brenner tumors occur between the fourth and the fifth decade of life, if benign, while the peak incidence is about 10 years later for borderline malignancy or malignant counterparts [1]. Proliferation of transitional (urothelial) type of epithelium among extensive fibrous stroma is a histopathological hallmark of the tumor. Hypothetically, it develops either from the ovarian surface epithelium or from uroepithelium improperly translocated during embryogenesis. Frequently, BTs contain components of other histopathological types of ovarian neoplasm, often mucinous. Less than 2% of all BTs reported in the literature were malignant or of low malignant potential [1].

Objective

The aim of the study was to analyze the case of a 74-year old woman operated at the Department of Gynecology, Gynecological Oncology and Gynecological Endocrinology of Medical University of Gdansk (POLAND) due to Brenner ovarian borderline tumor (BBT). Unexpectedly, recurrence occurred 25 months after the primary intervention, manifesting as a tumor in the abdominal wall.

Case report

A 74-year-old female (gravida 2, para 2) was admitted to our department in February 2011 with abdominal pain complaints for 5 months and a suspicion of a pelvic tumor of unknown biology. Transvaginal ultrasonography revealed a heterogeneous, complex (cystic-solid) tumor of the right ovary, 60x58mm in size. Serum Ca125 level was 34.2U/ml. Surgery revealed a cystic-solid tumor

of the right ovary, 7cm in diameter and with smooth external surface. The body of the uterus and the contralateral ovary corresponded to patient age and were macroscopically unchanged, so were other abdominal organs. Total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed.

Pathological examination revealed the tumor to measure 110x70x60mm. It was creamy-yellow and solid-cystic on the section, composed of few smooth cysts up to 2cm in diameter, filled with yellowish serous fluid. The tumor was originally diagnosed as benign Brenner tumor of the right ovary.

In April 2013, the patient presented at another hospital due to a subcutaneous tumor in the postoperative scar in the abdominal wall. Computed tomography (CT) scans of the abdomen revealed infiltrative lesion measuring 100x80x45 mm, located medially above the pubic symphysis. The tumor was resected radically and was diagnosed as Brenner tumor of borderline malignancy (BBT). Therefore, the original tumor was verified (prof. Wojciech Biernat) and finally diagnosed as BBT [Fig. XX]. The subcutaneous tumor in the postoperative scar was therefore diagnosed as a recurrence of BBT. The patient was informed about the possibility of complementary surgical procedure of omentectomy and lymph node biopsy, but in the meantime she died in September 2013 due to unknown causes.

Discussion

To the best of our knowledge, this is the first case of BBT recurrence in the postoperative scar of the abdomen described in the literature. There are only a few reports concerning Brenner tumors and even fewer about Brenner tumors of low malignant potential. Uzan et al., created the largest retrospective series of 35 patients with ovarian BBT so far [2]. The tumor was unilateral in 10 patients, similarly to our patient. There was one lethal recurrence (50 months after surgery) among the 5 patients involved in the follow-up. The authors described the recurrence as the first reported in literature. On the basis of their study, BBT could be regarded as a neoplasm with favorable prognosis but certainly many more cases are required to confirm it. Dierickx et al., tried to find some characteristic features of Brenner tumors

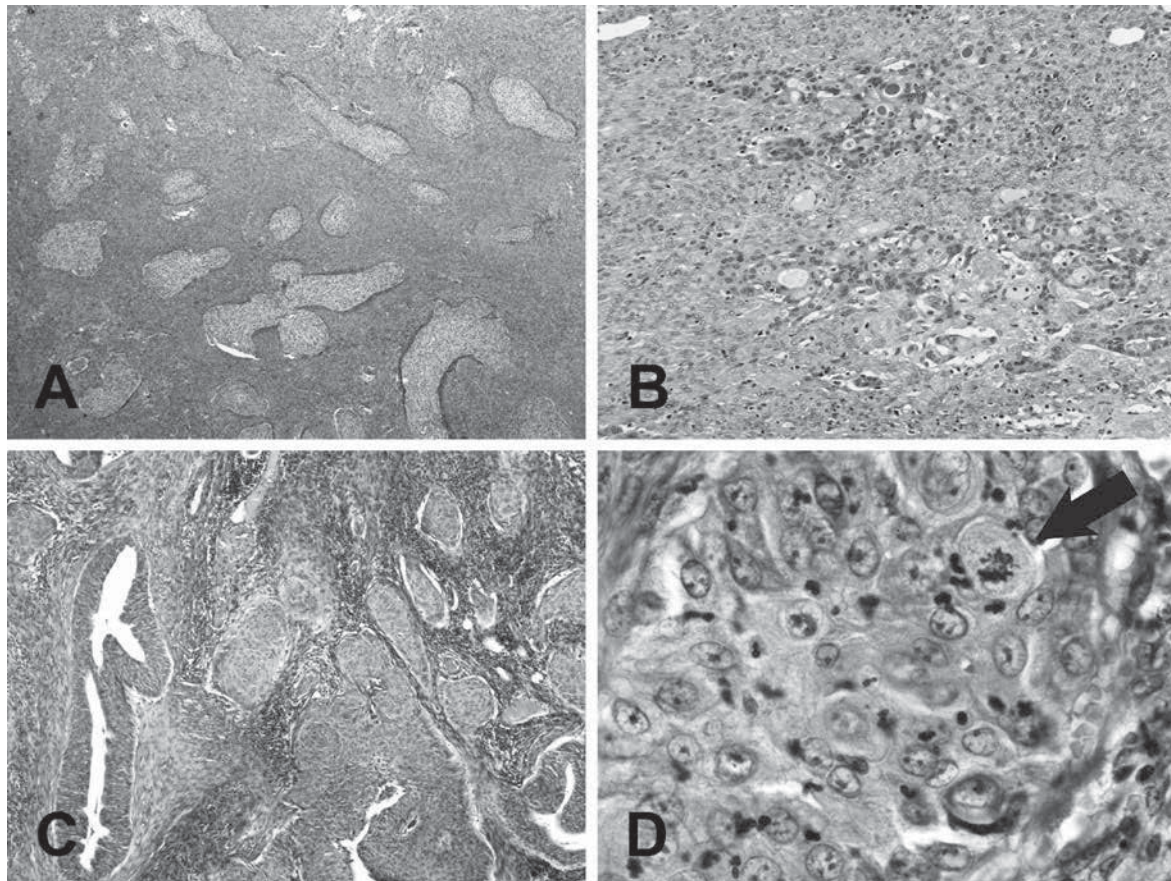


Figure 1. Ovarian borderline Brenner tumor (A,B). The tumor is composed of epithelial islands of tumor cells varying in size and shape randomly dispersed in the fibrotic stroma (A). Smaller nests infiltrating the background may signify more aggressive course of the tumor, however they do not present an overt atypia required for the diagnosis of malignant variant (B). The abdominal recurrence of the tumor (C,D). The cellular composition of the neoplasm is similar to the ovarian tumor. The epithelial component shows focal cystic (left part of the illustration) and prevailing solid type of growth (C). Higher magnification (D) discloses increased cellular atypia and individual atypical mitosis (arrow).

in ultrasound imaging [3]. At the basis of 29 cases of BTs (24 benign, 2 borderline and 3 malignant), they discovered that the majority of benign tumors on ultrasonography examination were solid, with weak or no Doppler signal, in contrast to borderline or malignant Brenner tumors which were cystic with strong Doppler signal. Thus, cystic-solid ultrasonography character of tumor in our case could suggest borderline tumor at least. Wang et al., tried to define some CT features typical for this neoplasm based on scans of 9 Brenner tumors, including one BBT [4]. Pure Brenner tumors, i.e. without other histopathological components, presented as a homogenous solid mass. As for additional components, they were usually cystic-solid or cystic lesions with calcifications on CT imaging. After contrast the density enhanced slightly in benign Brenner tumors, while in BBT the density increased highly on CT. On magnetic resonance imaging (MRI), benign BTs also differed from malignant ones, as the latter presented higher intensity in T-2 sequences [5]. Therefore, CT and MRI can be helpful in preoperative differentiation between benign BT and BBT. Positron emission tomography can differ benign and malignant ovarian neoplasm, but may be false negative in borderline tumors [6].

Malignant Brenner tumor metastasizes to the skin, lungs, central nervous system or shows tendency for intraperitoneal dissemination [7-9]. We were not able find a publication showing

similar behavior of BBT. Skin metastases from ovarian tumors occur rarely. Alcaraz et al., suggested that a wide range (0.6 to 10.4%) of all patients with malignant neoplasms of any organ develop skin metastases. In females, cutaneous metastases most commonly derive from the following primaries, in decreasing frequency: mammary carcinomas, ovarian, oral cavity, pulmonary, and large intestine [10]. Mean survival after the development of skin metastases is a few months [10]. Cormio et al., described nine patients with skin metastases from their own files of 220 patients with ovarian cancer [11]. Serous cancer dominated (7 cases out of 9); 5 patients were classified as FIGO III, 2 as FIGO I and 2 as FIGO IV. One patient presented skin metastases at the time of diagnosis of ovarian cancer, whereas in the rest mean metastases-free survival (MFS) was 23.4 months (range 4-37 months). Skin lesions were multifocal in the majority of cases, measuring from 0.5 to 3 cm. On follow-up, 8 patients died from disease progression and mean time to death since the development of skin metastasis was 4 months (range 2-65 months).

Our patient showed cutaneous involvement 25 months after ovarian resection. The secondary lesion was single and large (100x80x45 mm). The patient died 5 months after the skin recurrence, probably from other causes than BBT. It is hard to compare our patient to patients described by Cormio et al., due to different tumor biology and malignant potential.

Lukasz Klasa et al. *Recurrence of Brenner ovary borderline tumor in the abdominal wall postoperative scar...*

In the case of our patient, it is also intriguing how this cutaneous involvement can be perceived. As the lesion recurred in the scar, and the primary tumor presented neoplastic foci in close vicinity to the ovary surface, an implantation of freely dispatched fragment of BBT may be an alternative to the truly metastatic pathway of dissemination. Although exceedingly rare in Brenner tumors, implantations is a well-acknowledged pattern of abdominal involvement in other ovarian neoplasms of surface-stromal group. We are thus convinced that this unusual clinical course in BBT should be reported to make surgeons and gynecologist alert to the potential risk in patients with this rare entity.

Conclusions

BBT is a very rare, usually accidentally diagnosed, epithelial ovarian neoplasm. To the best of our knowledge, this is the first report of BBT skin recurrence in the literature. Due to rareness of such diagnosis and no statistical data bases, prognosis in BBT is truly hard to anticipate. According to the biggest BBT research of 35 cases with one lethal recurrence, it is possible to state that the prognosis is relatively favorable [2]. However, the literature lacks information on the prognosis of skin recurrence in BBT. Bad prognosis in skin metastases of ovarian cancer cannot be related to BBT metastases because of different histopathological diagnosis [11]. Some specific features on ultrasonography, CT and MRI described in the literature, could be helpful in preoperative differentiation between BBT and malignant/borderline tumor [3, 4, 5].

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