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Case report

Ovarian cystic tumor composed of brenner tumor and stroma ovarii

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ABSTRACT

Introduction: Brenner's tumors are transitional cell ovarian tumors composed of mature cells similar to urothelial cells forming nests within a fibromatous stroma.
Case report: In this observation we report the case of a brenner's tumor diagnosed in a 63 year old female patient. The positive diagnosis was difficult to retain.
Discussion: Brenner tumors are rare fibroepithelial ovarian tumors, representing 1 to 2 % of all ovarian tumors. They are almost always benign.
Conclusion: The treatment is essentially surgical and the indication of chemotherapy remains debatable.

1. Introduction

Brenner's tumors are transitional cell ovarian tumors composed of mature cells similar to urothelial cells forming nests within a fibromatous stroma. It was first described in 1898 by MacNaughton-Jones.

These tumors can be benign, borderline or malignant. Malignant brenner's tumors are exceptional which makes their therapeutic management difficult due to their extreme rarity and their prognosis considered to be poor [1]. All our work was reported in accordance with the SCARE criteria and guidelines [11].

2. Observation

The patient was 63 years old, without any particular pathological history, and the history of her disease goes back to 8 months with the appearance of pelvic pain of heaviness type without other associated signs. The clinical examination showed: on pelvic and endovaginal ultrasound: heterogeneous echostructure mass of the right flank measuring 124 × 82 mm and on abdomino-pelvic CT scan (Figs. 1 and 2): solid cystic and fatty itra-peritoneal mitotic lesion of 9.5 × 8.4 cm right para-umbilical without epenetration.

CA125: 16.82 U/ml and FCU: no sign of malignancy.

The patient was operated on by our team at the IBN ROCHED University Hospital in CASABLANCA, where she underwent a resection of the tumor (Fig. 3) and an appendectomy. The operation lasted 1 h without any bleeding complications and the postoperative course was simple.

Pathological examination was in favor of a poorly differentiated and invasive carcinoma of the right ovary, immediately suggestive of a malignant Brenner tumor (Fig. 4). Additional immunohistochemistry study showed negativity of GATA3 and WT1 and positivity of CK7: ovarian localization of a poorly differentiated carcinomatous proliferation.

6 months after the patient died.

3. Discussion

Brenner tumors are rare fibroepithelial ovarian tumors, representing 1 to 2 % of all ovarian tumors. They are almost always benign, making the diagnosis, prognosis and treatment of benign Brenner tumors well codified. However, the rarity of the malignant and proliferative variant (3 to 5 % of Brenner tumors) is the cause of diagnostic and therapeutic difficulties [2].

Brenner's tumors occur most often between the fourth and sixth decade, as in our patient's case.

Brenner tumors do not have characteristic symptoms. The clinical signs are not very specific, pelvic pain is generally in the foreground associated or not with a pelvic mass, abnormal vaginal bleeding or irregularity of the menstrual cycle can also be observed. They can also be revealed by an acute surgical complication such as torsion or

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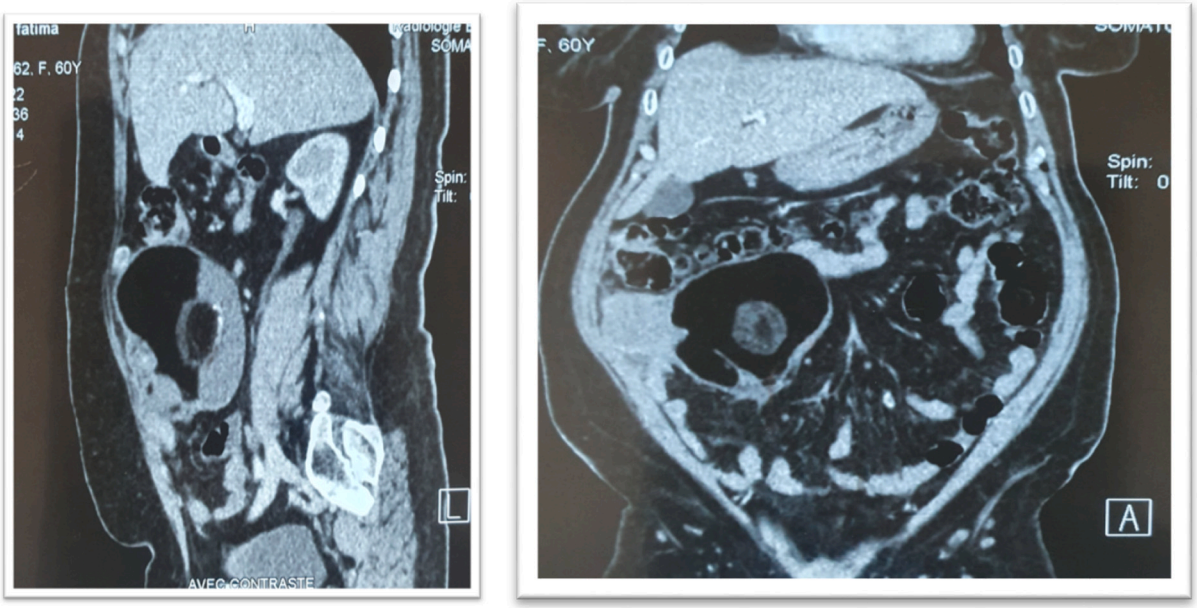

Figs. 1 and 2. Abdomino-pelvic scannographic image: solidoliquid heterogeneous cyst.

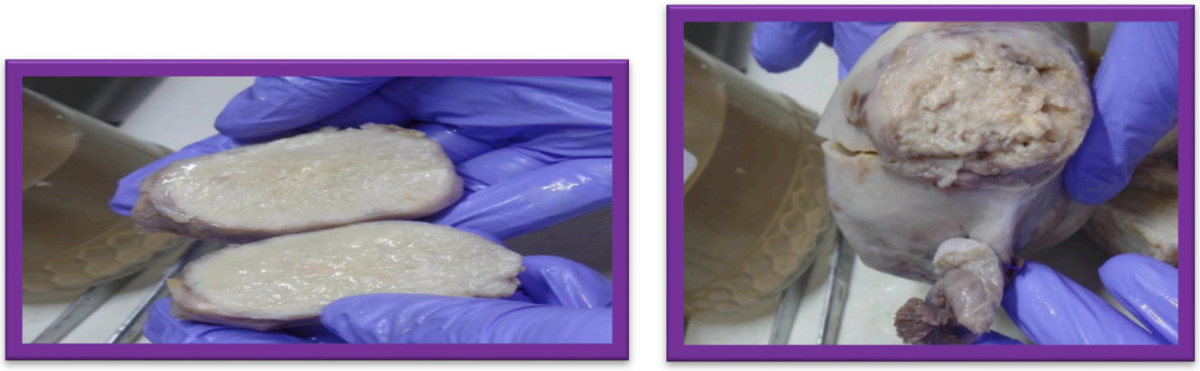

Fig. 3. Macroscopic image of a brenner tumor.

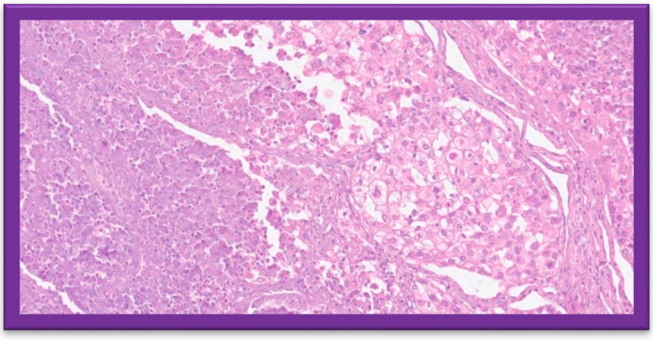

Fig. 4. Anatomopathological examination.
Poorly differentiated and invasive carcinoma of the right ovary, suggesting in the first place a malignant Brenner's tumor.

haemorrhage thus requiring an emergency treatment [3].

Some Brenner tumors have endocrine activity and secrete, most often, estrogen, responsible for metrorrhagia and endometrial hyperplasia. Rare cases have been reported to have androgenic activity with signs of virilization [4].

The most important serum marker in the evaluation of ovarian tumors is CA125, and CA19.9 and CEA are frequently combined. Many studies have shown that CA125 is neither sensitive nor specific enough to be an effective screening test. CA125 levels can be elevated in many pathological situations such as endometriosis, genital infections and even during ovulation. However, the CA125 assay is still of value in presuming malignancy when the level is elevated [5].

On ultrasonography, it is usually an echogenic mass, with an echogenicity close to the myometrium. It may contain areas of cystic transformation. The presence of large calcifications may wrongly lead to the diagnosis of subserous myoma with poor vascularity on color Doppler.

Hemorrhagic and necrotic changes are often seen in malignant Brenner tumors (Fig. 4).

MRI is not systematically requested. It is useful in solidolithic heterogeneous cysts. It can contribute to the preoperative diagnosis of malignancy in complex cysts and at the same time allows the assessment of locoregional extension in case of proven malignancy [5].

Macroscopically, they are most often unilateral, large, 20 cm in diameter, and greyish-white in appearance. Bilateral disease is uncommon, occurring in 12 % of cases. They are usually characterized by the presence of a solid component corresponding to the benign Brenner tumor associated with cysts containing papillary or polypoid masses [7].

Histologically, malignant Brenner's tumor is characterized by the

presence of typical foci of high-grade, rarely low-grade transitional cell carcinoma associated with a benign or borderline Brenner's tumor component embedded in a fibromatous stroma. Prominent glandular elements or calcification foci may also be associated. Extensive sampling of the tumor is necessary to establish the exact histological diagnosis [7].

Some authors reserve the term proliferative Brenner's tumor for tumors that mimic the appearance of a grade I urothelial carcinoma and consider the resemblance to grade II and III urothelial carcinomas as borderline [4,8]. The new WHO classification separates transitional cell carcinomas from Brenner tumors. In fact, the diagnosis of primary ovarian transitional tumors is made in the absence of a benign or borderline Brenner's contingent [8].

Malignancy [8].

The treatment of ovarian cysts is primarily conservative (cystectomy or oophorectomy), especially in a young woman, our patient benefited from a left adnexectomy, omontectomy and multiple biopsies as the criteria for benignity were met and extemporaneous examination was not available. However, in case of suspected malignancy, the treatment is radical; it consists of hysterectomy with bilateral adnexectomy. Laparoscopy remains the surgical technique of reference for presumed benign organic cysts. It has many advantages over laparotomy [6].

Concerning medical treatment, the use of adjuvant and even less neoadjuvant chemotherapy is controversial according to the authors, because of the low objective and histological response rates reported in the literature.

histological responses reported in the literature. Histological complete responses are exceptional and have been observed mainly after platinum-based multidrug therapy [9].

platinum-based multidrug therapy [9]. A standard paclitaxel-carboplatin regimen would be strongly recommended [10].

Benign Brenner tumors have an excellent prognosis, whereas malignant tumors remain of poor prognosis despite all therapeutic investigations. However, the prognosis of proliferating Brenner tumors seems to be excellent despite the small number of published cases and no deaths are reported in the literature [2,4,8].

Our patient underwent, in addition to radical treatment, an omentectomy with appendectomy.

4. Conclusion

Brenner's tumors represent only 1 to 2 % of all ovarian tumors. They are almost always benign and the rarity of the malignant variant is at the origin of diagnostic and therapeutic difficulties.

Anatomically, the characteristics of borderline and malignant tumors are not yet clearly defined as those of benign brenner tumors.

The treatment is essentially surgical and the indication of chemotherapy remains debatable.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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.

Ethical approval

I declare on my honor that the ethical approval has been exempted by my establishment.

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Declaration of competing interest

The authors declare having no conflicts of interest for this article.

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