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A case of ovarian psammocarcinoma with homolateral serous cystoadenofibroma and thecoma associated with Brenner tumour and cystoadenofibroma of the contralateral ovary

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Summary

Psammocarcinoma of the ovary is a rare serous neoplasm, with only 32 cases reported in the international literature. Characteristically, this tumour shows extensive formation of psammoma bodies, low-grade cytological features, and invasion of the ovarian stroma, peritoneum or intraperitoneal viscera. The behaviour of this entity is unpredictable, with benign, low malignant and metastatic potential. Herein the authors report a case of psammocarcinoma of the ovary with homolateral serous cystoadenofibroma and thecoma, which were associated with Brenner tumour and adenofibroma of the contralateral ovary, in a 78-year-old woman. Thus, this example shows an unpredictable tumour associated with multiple benign epithelial neoplasms and a benign stromal tumour. Moreover, this example of psammocarcinoma is very interesting because it measures only 1.5 x 0.5 x 1.5 cm and, to the best of the author's knowledge, represents the smallest case of psammocarcinoma described so far in the literature.

Key words: Serous psammocarcinoma; Serous cystoadenofibroma; Thecoma and Brenner tumour.

Introduction

Serous psammocarcinoma is a rare form of ovarian carcinoma with only 32 cases reported in the literature [1]. This neoplasm was first described by Gilks *et al.* as a lesion with massive psammoma body formation. The other histological criteria suggested by Gilks *et al.* for diagnosis of psammocarcinoma were as follows: (a) destructive invasion of ovarian stroma, vascular invasion, or invasion of intraperitoneal viscera; (b) moderate nuclear atypia; (c) presence of nests of solid epithelial proliferations no greater than 15 cells in diameter; (d) psammoma bodies that replace at least 75% of the papillae [2].

Here the authors report a further case of psammocarcinoma of the right ovary with homolateral serous cystoadenofibroma and thecoma which was associated with Brenner tumour and cystoadenofibroma of the contralateral ovary.

Case Report

A 78-year-old nulliparous woman, with a previous history of hypertension, and a subtotal hysterectomy as a result of severe uterine bleeding and leiomyomas at the age of 29 years, was admitted to the present institution to have her uterine cervix removed. Seric levels of Ca 125, Ca 19.9, and CEA were normal. A preoperative pelvic magnetic resonance image (MRI) revealed the presence of a right hand mass measuring 3.5 x 3 cm. This lesion was solid with a cystic area. A small cystic lesion was present in the left ovary: the patient underwent bilateral salpingo-oophorectomy.

The surgical specimens were fixed in ten percent neutral-buffered formalin for a routine light microscopic examination. Sections of neoplasms were submitted to histological examination and the samples were embedded in paraffin and stained with haematoxylin-eosin.

On sectioning, the right ovary disclosed the presence of three lesions. Two of these were solid, while the third was cystic. One of the solid lesions was a small, heavily calcified, grey sub-capsular nodule measuring 1.5 x 0.5 x 1.5 cm (Figure 1).

Histologically, this small nodule corresponded to a psammocarcinoma and was characterized by numerous psammoma bodies (Figure 2A) which were occasionally surrounded by papillary and tubular structures, lined by cytological bland cuboidal or low columnar epithelium (Figure 2B).

The cystic neoplasm macroscopically contained serous fluid and papillary projections. Histologically, this neoplasm corresponded to a serous cystoadenofibroma which, characteristically, had formed broad papillae which projected into the lumen of a cyst. Both these papillae and the wall of the cyst showed cellular fibrous stroma and were lined by a single layer of columnar epithelium without nuclear atypia (Figure 3A). The other solid lesion appeared as a mass measuring 3 x 1.5 cm, with a yellow sectioned surface (Figure 1A). Microscopically, this mass revealed the features of thecoma showing a fibromatous background with spindle cells and masses of cells which had abundant pale cytoplasm and oval/round nuclei (Figure 3B).

Macroscopically, the left ovary revealed the presence of another epithelial neoplasm which corresponded to a benign Brenner tumour, showing the presence of epithelial nests with cystic structures containing eosinophilic debris (Figure 4A). The epithelial elements were characterized by eosinophilic cytoplasm and oval nuclei with small nucleoli and longitudinal grooves (coffee bean nuclei) (Figure 4B). In addition, in other areas of the same ovary, there was a small cyst measuring one by one cm into which projected some papillary excrescences which microscopically revealed the histological features of cystoadenofibroma (Figure 4C). After 48 months from the diagnosis, the patient is alive without evidence of disease.

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Fig. 1

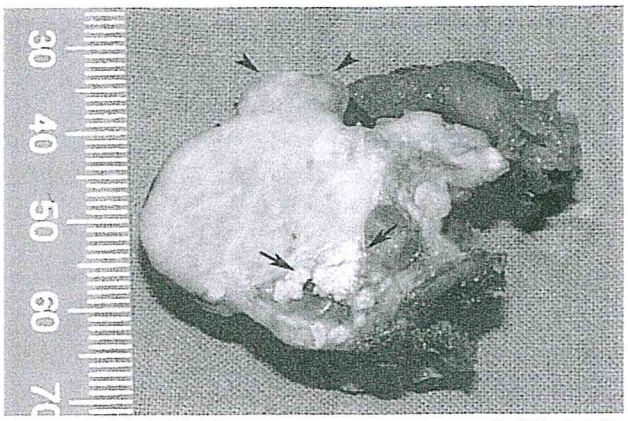

Fig. 3A

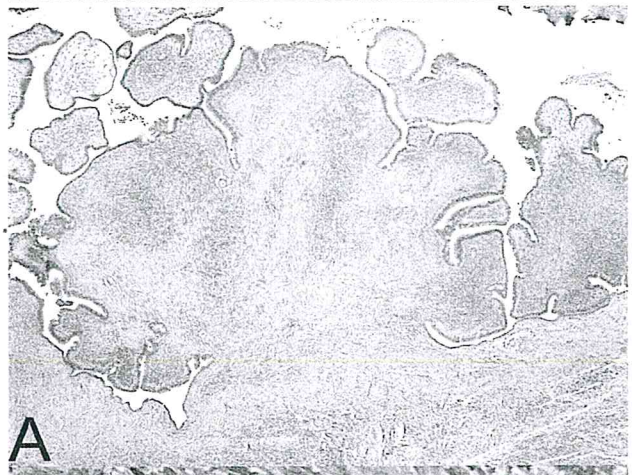

Fig. 3B

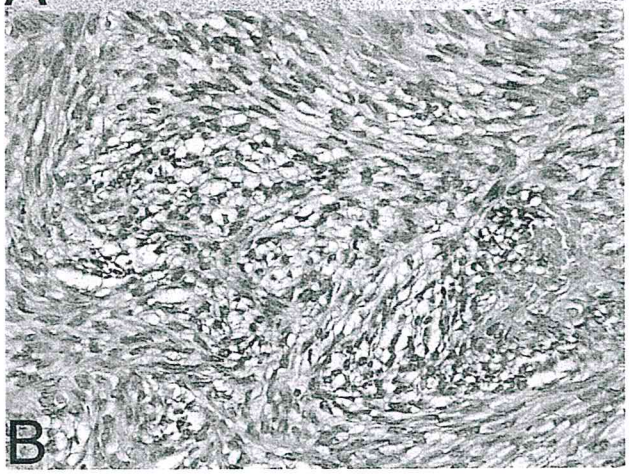

Fig. 2A

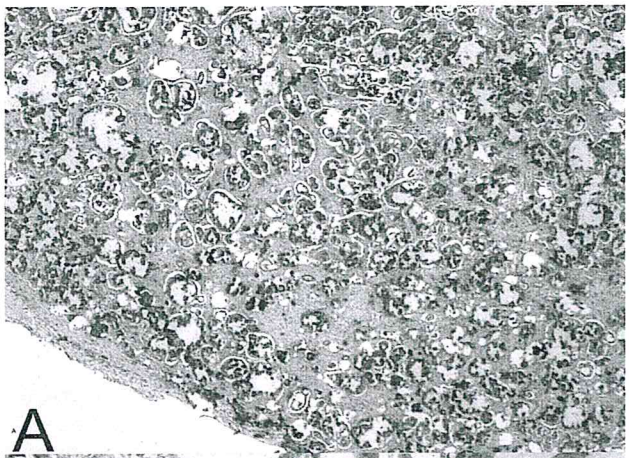

Fig. 2B

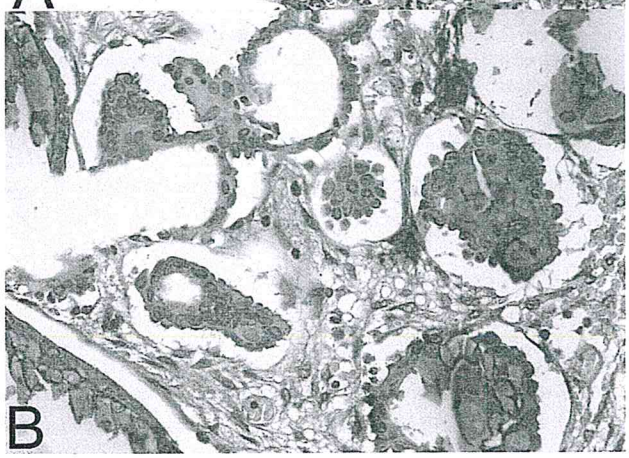

Figure 1. — Right salpingo-oophorectomy specimen containing a sub-capsular, small calcified nodule (arrow heads). A): a solid yellow mass and a cystic lesion with papillary structures (arrows).
Figure 2. — On histological examination, the sub-capsular calcified lesion corresponds to a psammocarcinoma with extensive psammoma bodies (A: haematoxylin-eosin x 40) which are surrounded by single layer of cytological bland cuboidal or low columnar epithelium (B: haematoxylin-eosin x 400).
Figure 3. — The cystic lesion shows features of serous cystoadenofibroma revealing the presence of broad papillae with cellular fibrous stroma and a single layer of epithelium projecting into the lumen of a cyst (haematoxylin-eosin x 100). The solid yellow mass corresponds to a thecoma showing a fibromatous background with spindle cells and masses of cells which have abundant pale cytoplasm and oval/round nuclei (e: haematoxylin-eosin x 200) haematoxylin-eosin x 40).

Discussion

Thecomas or theca cell tumours are benign ovarian neoplasms composed only of theca cells. Histogenetically, they are classified as sex cord stromal tumours. They are typically estrogen-producing and occur in older women (mean age 59 years; 84% after menopause). They can, however, appear before menopause [3].

Sixty percent of patients present with abnormal uterine bleeding, and 20% have an endometrial carcinoma.

Grossly, the tumour is solid and yellow. Microscopically, the tumour cells have pale, abundant, and lipid-filled cytoplasms.

Ovarian cystoadenofibroma are infrequent superficial epithelial tumours. They can appear at all ages. Macroscopically, it has been seen that in younger patients, they are cystic with small papillae, while in older patients they form fibrotic nodules.

Brenner tumours are uncommon surface-epithelial stromal cell tumours They are most frequently found as incidental findings on pelvic examination or at laparotomy [4]. These tumours may be very small to very large, and may be solid or cystic.

Its epithelial cell (which defines these tumours) is a transitional cell and similar in appearance to bladder

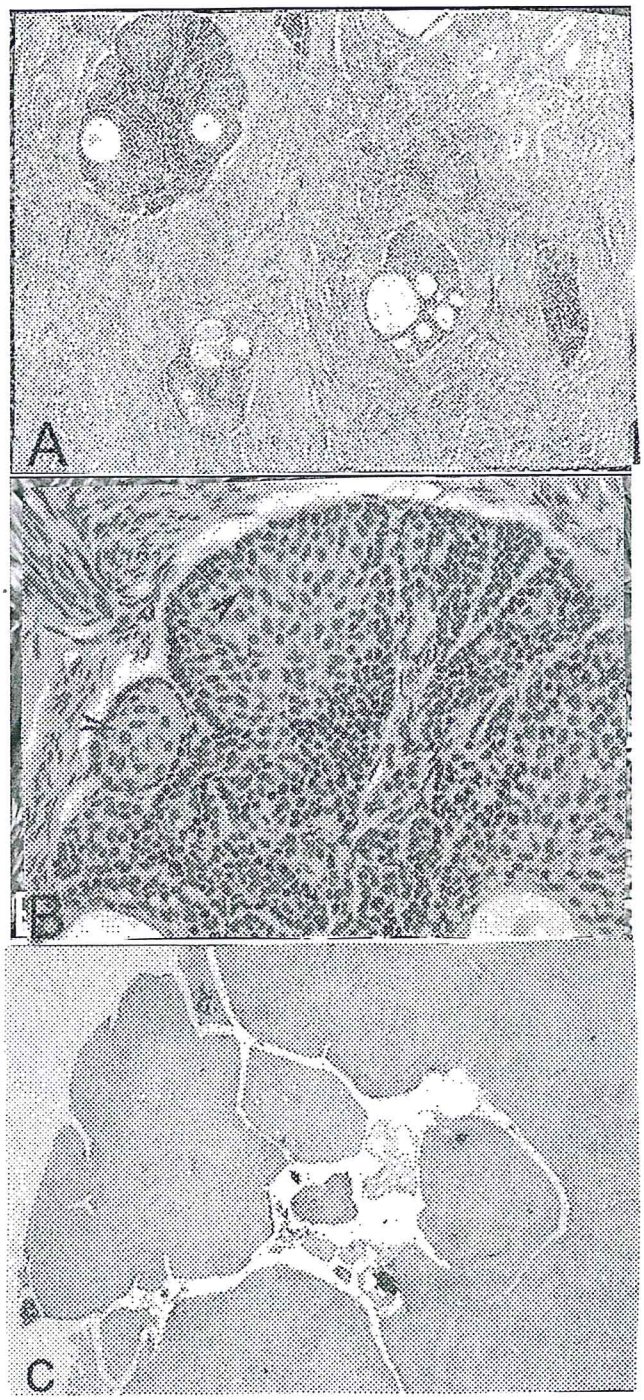

Figure 4. — On histological examination, the left ovary reveals the presence of two benign neoplastic lesions: a Brenner tumour which is composed by tissue that resembled normal ovarian fibrous tissue and nests of transitional cells (A: haematoxylin-eosin x 100) with nuclear longitudinal grooves (coffee bean nuclei) (B: haematoxylin-eosin x 400, arrow heads: nuclear longitudinal grooves) and a serous cystoadenofibroma (C: haematoxylin-eosin x 40).

epithelium. The nests of transitional cells are surrounded by tissue that resembles normal ovary and their nuclei present longitudinal grooves (coffee bean nuclei).

As a rule, the neoplasms which can be associated with Brenner tumour have already been described and they can be epithelial, germinal or sometimes ovarian stromal tumours [5-7].

A review of the literature reveals that psammocarcinoma of the ovary can be associated with cystoadenofibroma [8], but was never observed with thecoma or other stromal ovarian tumors.

In the present case, the authors observed the coexistence of different types of epithelial neoplasms and a stromal tumour associated with a psammocarcinoma, which can be considered a malignant neoplasm.

Moreover, this example of psammocarcinoma is very interesting because it measures only 1.5 x 0.5 x 1.5 cm and, to the best of the author's knowledge, represents the smallest example of psammocarcinoma described so far in the literature [1].

In the authors' opinion, because psammocarcinoma is characterized by unpredictable behaviour with both low malignant, aggressive [9], and metastatic potential [10], accurate follow-up is always mandatory.

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