Chemotherapy in the Treatment of Ovarian Psammocarcinoma: A Case Report and Review of the Literature

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Abstract

Introduction: Psammocarcinoma is a rare form of epithelial serous ovarian carcinoma characterized by extensive formation of psammoma bodies, invasion of ovarian stroma, peritoneum or intraperitoneal viscera, and moderate cytological atypia. These tumors represent a real problem of the diagnostic, and the role of chemotherapy is not yet clearly demonstrated.

Case presentation: We herein report a case of psammocarcinoma of ovary with peritoneal carcinosis in a forty year old Moroccan female. The patient underwent optimal surgical debulking and nine courses of chemotherapy with carboplatinum and paclitaxel with a complete response. The prognosis for this type of ovarian cancer is unclear, but it appears to be better than other forms of epithelial ovarian cancer.

Conclusion: The psammocarcinome is a rare entity, majority of patients are diagnosed at an advanced stage. The role of chemotherapy is poorly defined; some authors have their patients treated by neoadjuvant or adjuvant chemotherapy.

We currently lack evidence of increasing the benefits that can bring chemotherapy in the management of advanced ovarian psammocarcinomas. Only trials in wide yard can answer this question.

Keywords: Psammocarcinoma; Ovarian; Complete response

Introduction

Psammocarcinoma is a rare form of epithelial serous ovarian carcinoma characterized by extensive formation of psammoma bodies, invasion of ovarian stroma, peritoneum or intraperitoneal viscera, and moderate cytological atypia [1]. These tumors represent a real diagnostic problem, of the fact that there are not clinical and pathological features that allow us to differentiate serous borderline [2].

There are no therapeutic standard guiding the management of psammocarcinomas in the advanced stages, and the role of chemotherapy is not yet clearly demonstrated.

The authors reported a case of an ovarian psammocarcinoma diagnosed at an advanced stage and was successfully treated only by chemotherapy.

Case Report

A 40 year old woman, diabetic, hypertensive under treatment for 10 years, its history back by the sudden onset of abdominal pain, diffuse pelvic evolving in a context of deterioration of general condition. The Physical examination in the admission reveals a patient with a performance status at 2. Pelvic examination showed a sensitive voluminous abdominopelvic mass with abundance ascitis. Abdomino pelvic computed tomography scan revealed a heavily calcified abdominopelvic mass with peritoneal carcinosis. The serum CA-125 level was elevated (400 UI/ml; normal value: <35 UI/ml). The patient underwent a surgical exploration which revealed generalized carcinosis.

The biopsy of the mass of the left ovary revealed a psammocarcinoma (Figure 1). She was then referred to our department for treatment. Primary chemotherapy was planned. The patient received three courses of paclitaxel 175 mg/m² and carboplatin (AUC 5).

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The CA125 level went from 400 to 50 IU, and we had a radiological stabilization with no other distant lesions. The patient has been proposed for revision surgery but because the radiological stabilization, and the good tolerability of the treatment, we decided to continue the chemotherapy. Evaluation after 9 cycles of chemotherapy objectified a complete clinical response and normalization of the level of CA125 and we had a radiological stabilization in most cases, a regression of the disease but the period of regression of the disease often diagnosed at an advanced stage, the results showed treated with neoadjuvant, adjuvant or consolidation chemotherapy with the disease often diagnosed at an advanced stage, the results showed that can bring chemotherapy in the management of advanced ovarian forms, however, we currently lack evidence of increasing the benefits and few cycles of chemotherapy in post operative in the aggressive biological response form of advanced ovarian psammocarcinoma. 

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### Discussion

The psammocarcinome is a rare entity that more frequently localized in the thyroid [3], Meninges [4] and at the extended gastrointestinal [5,6]. Its localization except ovarian and limited to a few cases reported in the literature [7-9]. Histological diagnosis is difficult, it uses criteria determined by morphological Gilks which should be sought as extensive training bodies of psammoma which appear calcified, rolled, invasion of ovarian stroma and a typical cytology moderate [7,10], it is the case of our patient in whom the diagnosis of ovarian psammocarcinoma could not be carried after a second reading. Majority of patients are diagnosed at an advanced stage as in the case series Gilks and other series [11-14]. Because of their rarity there are no specific recommendations treatment, most authors recommend aggressive cytoreduction [15]. The role of chemotherapy is poorly defined; some authors have their patients treated by neoadjuvant or adjuvant chemotherapy. The care of patients who cannot benefit from surgical treatment to be defined in particular the role of chemotherapy.

Kelley et al. [11] reported in their work (Table 2) series of 12 cases treated by surgery or surgery and chemotherapy and conclude a possible benefit of adjuvant chemotherapy in aggressive forms.

By referring to the treatment of peritoneal psamocarconomes is described in the literature including different sets of patients were treated with neoadjuvant, adjuvant or consolidation chemotherapy with the disease often diagnosed at an advanced stage, the results showed in most cases, a regression of the disease but the period of regression varied from one case al depending on the type of surgery done to the chemotherapy regimen adopted as shown (Table 3).

There is no limit to our research of cases treated with chemotherapy alone, the case report was distinguished by an excellent clinical and biological response form of advanced ovarian psammocarcinoma.

### Conclusion

The standard treatment of ovarian psammocarcinoma is surgery and few cycles of chemotherapy in post operative in the aggressive forms, however, we currently lack evidence of increasing the benefits that can bring chemotherapy in the management of advanced ovarian

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### Table 2: Ovarian psammocarcinoma clinical features [16].

| Case | Age (years) | Treatment | FIGO stage | Follow up |
|------|-------------|-----------|------------|-----------|
| 1    | 66          | LSO       | IIIB       | LFU       |
| 2    | 72          | BSO       | IIIA       | NED       |
| 3    | 55          | TAH/BSO chemo | IIIA | NED     |
| 4    | 36          | TAH/BSO   | IIIB       | NED       |
| 5    | 53          | TAH/BSO omentectomy | IIIB | NED     |
| 6    | 53          | TAH/BSO omentectomy | IIIB | LFU     |
| 7    | 76          | BSO omentectomy | IIIB       | NED       |
| 8    | 59          | LSO       | IIIIC      | DOD       |
| 9    | 18          | Optimal debulking, chemotherapy | IIIIC | NED     |
| 10   | 49          | BSO       | IA         | NED       |
| 11   | 59          | TAH/BSO omentectomy, appendectomy bilateral pelvic and peri-aortic lymphadectomy | IIIB       | NED       |
| 12   | 48          | TAH/BSO omentectomy | IIIA       | NED       |

LFU: Lost to Follow Up; NED: No Evidence of Disease; DOD: Dead of Disease

TAH: Total Abdominal Hysterectomy; BSO: Bilateral Salpingo Oophorectomy

### Table 3: Peritoneal serous: clinical features.

| Authors | Age | Presenting symptoms or signs | Surgery | Chemotherapy/ radiotherapy | FIGO stage | Follow up |
|---------|-----|------------------------------|---------|----------------------------|------------|-----------|
| Gilks et al. [10] | 58 | Abdominal swelling | Supracervical hysterectomy, BSO | NA | IIIB | INF 1 year |
| Gilks et al. [10] | 55 | Abdominal pap smear | TAH/BSO omentectomy | NA | IIIA | NED: 10 years |
| Gilks et al. [10] | 48 | Menometrorhagia, pelvic mass | TAH/BSO omentectomy | NA | IIIB | INF 1 year |
| Molpus et al. [14] | 58 | Increasing abdominal girth, progressive, shortness of breath | TAH/BSO omentectomy debuking | None | IIIIC | PFS: 4 years |
| Munkarah et al. [8] | 27 | Abdominal pain | Conservatrice surgery, secondary debuking after chemotherapy | Cyclophosphamisec/ cisplatine 6 cycles | IIIC | NED: 6.5 years |
| Weir et al. [9] | Report of 7 patients | Average 48 years (range 42-72 years) | Pelvic mass (in 3/7 patients) | 2 patients TAH/BSO omentectomy | One patient: taxol– carboplatine | No data | Data were available in 3 out of 7 cases NED at 1,4,3,8,8,3 years |

TAH: Total Abdominal Hysterectomy; BSO: Bilateral Salpingo Oophorectomy; LSO: Left Salpingo Oophorectomy; USO: Unilateral Salpingo Oophorectomy; BSO: Bilateral Salpingo Oophorectomy; NED: No Evidence of Disease; DOD: Dead Of Disease; PFS: Progression Free Survival; AWD: Alive With Disease
psammocarcinomas. Only trials in wide yard can answer this question.

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