

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).

Evaluation after 3 cycles showed a net clinical benefit with radiological stabilisation in the Chest Abdominal and pelvic CT scan. The decision was to continue the same chemotherapy.

The patient received three other courses, evaluation showed a net clinical benefit with a performance status at 1, the disappearance of the pain and drainage of ascitis.

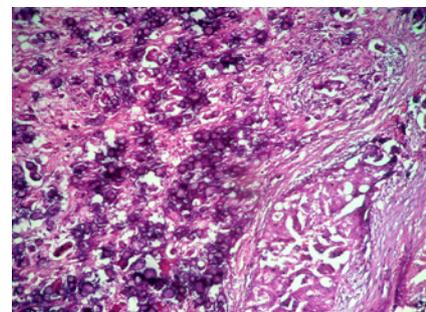


Figure 1: Histological examination: numerous psammomas bodies with tumoral cells right down (HE×10).

Rates of CA125	CA125 (UI /ml)
Before chemotherapy	400
After 6 cycles of chemotherapy	50
After 9 cycles de chemotherapy	28

Table 1: Rates of CA12 5 before and after treatment.

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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	IIIC	DOD
9	18	Optimal debulking, chemotherapy	IIIC	NED
10	49	BSO	IA	NED
11	59	TAH BSO omentectomy, appendectomy bilateral pelvic and periaortic lymphadenectomy	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 2: Ovarian psammocarcinoma clinical features [16].

Authors	Age	Presenting symptoms or signs	Surgery	Chemotherapy/radiotherapy	FIGO stage	Follow up
Gliks et al. [10]	58	Abdominal swelling	Supracervical hysterectomy, BSO	NA	IIIB	INF 1 year
Gliks et al. [10]	55	Abdominal pap smear	TAH/BSO omentectomy	NA	IIIA	NED: 10 years
Gliks et al. [10]	48	Menometrorrhagia, 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 debulking	None	IIIC	PFS: 4 years
Munkarah et al. [8]	27	Abdominal pain	Conservative surgery, secondary debulking after chemotherapy	Cyclophosphamide/cisplatin 6 cycles	IIIC	NED: 6,5 years
Weir et al. [9] Report of 7 patients	Average 48 years (range 42-72 years)	Incidental (in 3/7 patients) Pelvic mass (in 3/7 patients) Abdominal pain (in 1/7 patients)	2 patients TAH/BSO omentectomy 2 patients: BSO, omentectomy 1 patient: USO, omentectomy 1 patient: myomectomy	One patient: taxol-carboplatin One patient: cisplatin-cyclophosphamide	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;
USO: Unilateral Salpingo Oophorectomy; LSO: Left Salpingo Oophorectomy;
NA: Not Administered; NED: No Evidence of Disease DOD: Dead Of Disease;
PFS: Progression Free Survival; AWD: Alive With Disease

Table 3: Peritoneal serous: clinical features.

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 (Table 1) but always radiological stabilization.

Discussion

The psammocarcinoma 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 psammocarcinomas 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 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

psammocarcinomas. Only trials in wide yard can answer this question.

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