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Case Report

Psammomatous ovarian tumor arising from the surface epithelium a borderline serous tumor

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Abstract

Psammocarcinoma is a rare variant of ovarian serous carcinoma and similar to carcinomas that exhibit low-malignant potential. A woman aged 38 years was admitted to the gynecology clinic with abdominal distension and amenorrhea. Magnetic resonance imaging (MRI) showed lobulated calcified abdomino-pelvic masses and multiple peritoneal implants. The patient was diagnosed as having ovarian tumor and underwent surgery. During the operation a papillary solid mass approximately 100x150 mm in diameter was found that involved the adnexa bilaterally, showed adhesions to the pelvic organs, and had totally invaded the uterus. In addition, multiple tumoral implants ranging from 2- 3 cm in size were seen on the bladder, in the anterior wall of the abdomen, and omentum. An optimal debulking surgery was performed. Pathology reported a borderline serous tumor in the left adnexal region, a microinvasive serous tumor and psammomatous carcinoma in the right adnexal region, and bilateral non-invasive desmoplastic implants in the pelvic lymph nodes. The patient underwent 6 cycles of paclitaxel and carboplatin chemotherapy after surgery. No recurrent tumor has been observed since the primary surgery in November 2014. This case report draws attention to the recommendation for surgery and subsequent administration of adjuvant chemotherapy in psammomatous ovarian cancer.

Key words:

Borderline serous tumor, psammocarcinoma, ovary

Introduction

Psammocarcinoma is a rare variant of ovarian serous carcinoma characterized by widespread psammoma body formation, and invasion of ovarian stroma, peritoneum, or intraperitoneal visceral organs [1,2]. As occurrence of the tumour is rare and there are no long-term follow-up data in the literature, the biologic behavior is not fully understood [3]. Although considered low-grade carcinomas, these tumors may be benign or malignant [4]. Only 32 cases of psammomacarcinoma have been reported in the literature to date [4]. Herein, a case of psammomatous ovarian tumor arising from the surface epithelium of a borderline serous ovarian tumor with extensive peritoneal and lymph node implants is presented.

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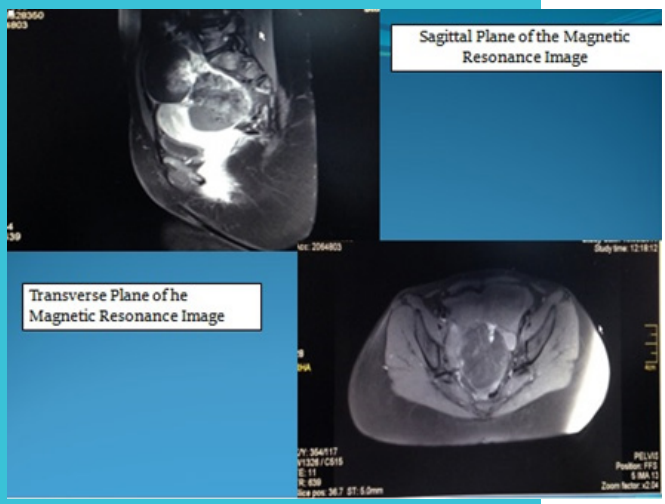
Case presentation

A gravida 3, parity 2, abortus 1 woman aged 38 years was admitted to our gynecology clinic with abdominal distension and amenorrhea. Vaginal ultrasonography revealed a uterus of normal dimensions but there was a 114x64 mm cystic structure that included solid fields in the right adnexal region, and a 97x70 mm mass arising from the left ovary that included mainly solid components. Ascites was also detected in the abdomen. The serum CA 12-5 level was 687 U/mL. Contrast-enhanced magnetic resonance imaging (MRI) showed lobulated calcified abdomino-pelvic masses and multiple peritoneal implants (Figure-1). The patient was diagnosed as having ovarian tumor and underwent surgery in November 2014. During the operation, we observed a papillary solid mass approximately 100x150 mm in diameter, which showed adhesions to the pelvic organs, involved bilateral adnexa, and was totally invasive to the uterus. Multiple tumoral implants ranging from 2 to 3 cm in size were seen on the

bladder, and in the anterior wall of the abdomen and omentum. Ascites was also present. An optimal debulking surgery was performed leaving a residual tumor of less than 1 cm. The histopathologic examination of the specimen reported a borderline serous tumor in the left adnexal region; in the right adnexal region there was a microinvasive borderline serous tumor and a psammomatous carcinoma had developed with bilateral non-invasive desmoplastic implants in the pelvic lymph nodes (Figure 2-3). The patient underwent 6 cycles of paclitaxel and carboplatin chemotherapy after the surgery. No recurrence of tumor has been observed since the primary surgery in November 2014 and the patient has been followed up intensively.

Figure 1.

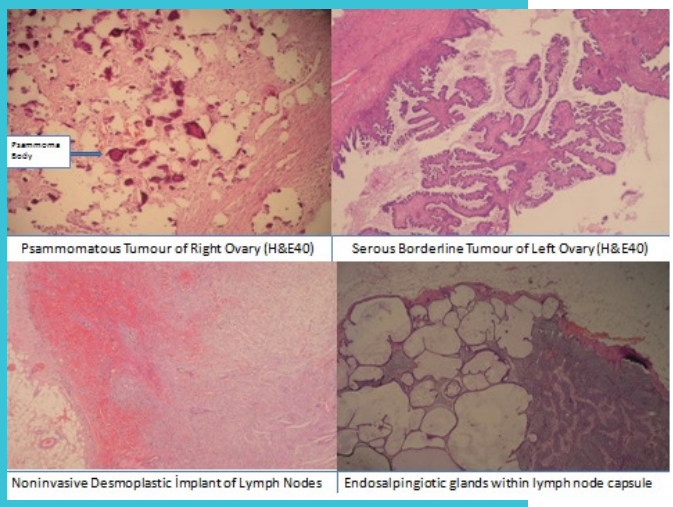


Sagittal and transverse plane of the magnetic resonance image

Discussion

Psammomatous carcinoma is an infrequent type of ovarian cancer [5]. Cases of psammomacarcinoma tumors have been reported in patients aged 18-76 years [5,6]. Although these patients were usually admitted with abdominal pain and swelling, 43% were incidentally diagnosed [7]. Histomorphologic criteria used for pathologic differential diagnosis were first determined by Gilks et al and revised by Chen et al [5, 8]. Serous tumors that may contain psammoma bodies are taken into consideration in the differential diagnosis of psammocarcinoma. Differentiation of psammocarcinoma from other tumor types must be made very carefully because there are differences in treatment and survival between psammocarcinoma and other tumor types [5]. High-grade serous adenocarcinomas usually exhibit higher cellular atypia.

Figure 2.

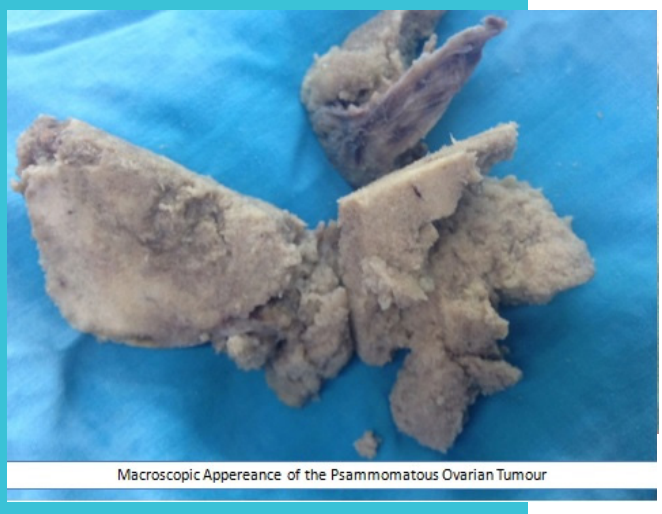


Psammomatous tumour of right ovary (H&E40); serous borderline tumor of left ovary (H&E40); noninvasive desmoplastic implants of lymph nodes; endosalpigiotic glands within lymph node capsule

Typical low-grade serous adenocarcinomas do not contain psammoma particles in more than 75% of papillae, which is a diagnostic criterion for psammocarcinoma. Unlike other serous tumors, serous psammocarcinomas do not form solid epithelial islands in more than 15 cells [5]. In order to make a diagnosis of psammocarcinoma with these criteria, there should be destructive invasion in the ovarian stroma or peritoneum, atypia must be at the most moderate level, the extent of epithelial cell groups must not exceed 15 cells, and at least 75% of papillae or epithelia should contain psammoma bodies. The current case was evaluated under these criteria and was diagnosed as psammocarcinoma. Cystadenofibroma psammocarcinoma should also be considered in the differential diagnosis. Some rare cystadenofibromas contain light cytologic atypia and psammoma particles [9]. However, cytologic atypia is relatively unclear in such lesions, and psammoma particles are not intense. Our case had severe cytolog-

ic atypia and contained intense psammoma particles. Serous borderline tumors and low-malignant potential tumors can be distinguished by the presence of destructive invasion of abdominal organs and the peritoneum [10]. The invasive nature psammocarcinomas differentiates them benign proliferations, such as endosalpingiosis. The moderate atypia of the tumor cells makes it possible to differentiate psammocarcinomas from high-grade serous carcinomas [11]. Magnetic resonance imaging (MRI) is the most commonly preferred imaging method as for all other types of ovarian cancers. The presence of diffuse sand-like calcifications in a T1-weighted MRI image with a pelvic mass should be considered as a radiologic marker for psammomatous carcinoma [12]. Dong A. et al. concluded that due to glucose metabolic activity, positron emission tomography combined with computed tomography (PET-CT) was advantageous for the diagnosis of malignant calcification and could be used to diagnose and stage serous psammocarcinoma of the ovary [13]. The MRI findings of the current patient indicated lobulated, scattered, calcified abdominopelvic masses and multiple peritoneal implants (Figure-1).

Figure 3.



Macroscopic Appereance of the Psammomatous Ovarian Tumour

Macroscopic appearance of the psammomatous ovarian tumour

These tumors often progress silently and are usually diagnosed at an advanced stage [3, 6, 12]. As in most cases in the literature, the tumor in the current case was identified at stage III. Even though such tumors present at an advanced stage with aggressive invasion, they have a low risk of metastasis and good clinical course. Thus, there are reports of patients who never received chemotherapy even though their tumors were at an advanced stage [14]. Although the survival rate of patients with these tumors has been reported to be high, there may occasionally be recurrence; there is only one documented psammocarcinoma-related death in the literature [5]. Norese G et al. reported a patient with stage IIIC psammocarcinoma of the ovary who underwent surgery in October 2004, and received 6 cycles of chemotherapy (paclitaxel & carboplatine). The tumor recurred in February 2005, and thus a secondary cyto-reductive surgery was performed in May 2005, followed by radiotherapy. During a long follow-up period, the tumor was not observed again until December 2014 [15]. Kelly et al. presented a patient aged 18 years with psammocarcinoma of the ovary (stage III) who underwent total abdominal hysterectomy, bilateral salpingoopherectomy and omentectomy, as well as postoperative chemotherapy. Disease-free survival was reported as 42 months [6]. Shah et al. presented a nulliparous patient aged 27 years who had had psammoma carcinoma of the peritoneum and achieved successful pregnancy following fertility-sparing surgery and adjuvant chemotherapy [16]. Grimaldi et al. recommend fertility-sparing surgery if a patient is young and no macroscopic tumoral implants are present on the ovaries [17]. Although the biologic behavior of these tumors is not precisely known, the general approach tends towards chemotherapy subsequent to debulking surgery [1, 5, 6, 12]. Our patient had widespread tumoral implants and also ascites in the abdomen (Stage IIIC); therefore, we opted for optimal debulking surgery and adjuvant chemotherapy. In this regard, the most frequently used method is a cytotoxic treatment comprising cyclophosphamide combined with tamoxifen, cisplatin or carboplatin [18]. In the current case report, a nulliparous woman aged 38 years presented with multiple intra-abdominal- and lymph node metastases. Although psammocarcinoma ovarian cancer is known to have low metastatic potential, the patient underwent surgery as a case of advanced-stage ovarian cancer because of the presence of diffuse intra-abdominal and lymphatic metastases. The patient was administered 6 cycles of adjuvant chemotherapy and has been closely monitored through examinations at specific intervals since surgery in November 2014. Even though it is still too early to report a survival rate for this case, no recurrence of tumor has been observed since the optimal debulking surgery performed in November 2014.

Psammomatous ovarian cancer is similar to carcinomas and exhibits low-malignant potential with a slow clinical course. The aim of this case study of a cancer with a multiple metastatic course was to draw attention to the recommendation for surgery and subsequent administration of adjuvant chemotherapy for psammomatous ovarian cancer.

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None

Conflict of Interest
The authors have no conflict of interest.

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