Case Report

Extensive peritoneal keratin granuloma in stage IV B endometrial carcinoma with an outstanding survival: A case report and review of the literature

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Introduction

Peritoneal keratin granuloma is a rare lesion included among reactive tumor-like lesions of the peritoneum. It can be secondary to endometrioid adenocarcinoma with squamous differentiation of the endometrium and ovary, atypical polypoid adenomyoma of endometrium and in association with ruptured dermoid cysts.

The prognostic significance of these lesions is unknown but it seems to have no interference in prognosis, when no viable metastatic cells are detected.

Here we describe a case of an endometrioid adenocarcinoma of the endometrium, in which diffuse peritoneal keratin granulomas were found with viable tumor implants and an outstanding survival.

Case report

A 69-year-old woman with preceding post-menopausal bleeding, was referred to our oncology center after an endometrial biopsy, revealed an endometrial endometrioid adenocarcinoma. Her past medical history only included medicated and controlled hypertension. She was proposed for surgical staging and during exploratory laparotomy multiple implants were found, suspicious of metastatic tumor in inferior abdomen (colon, small bowel, mesentery) and pelvis (pouch of Douglas and vesicouterine pouch, leading to a very adherent uterus). Surgery consisted of peritoneal fluid removal and biopsy of two implants. Histopathologic analysis revealed an intense inflammatory process with foreign body giant cells resembling peritoneal keratin granulomas (Figs. 1 and 2). Peritoneal fluid was negative for tumor cells. With no history of granulomatous diseases, the diagnosis was questioned, leading to a revision of the endometrial biopsy which reassured former diagnosis of endometrioid adenocarcinoma. It was decided to perform a new surgery, which revealed similar findings, with intraoperative frozen sections. Surgery consisted only of biopsy of a nodule from the peritoneal surface and another from the omentum because intraoperative frozen sections, confirmed the presence of viable tumor implants of poor differentiated adenocarcinoma immunostaining, for cytokeratin (MNF) at the peritoneal nodule and peritoneal keratin granulomas in the omentum one.

With the diagnosis of endometrial endometrioid adenocarcinoma, Fédération Internationale de Gynécologie et d’Obstérétique (FIGO) stage IV B, she was submitted to utero-vaginal brachytherapy (two applications of 137Cs) and started megestrol acetate 160 mg/day during 2 years. Twelve years after the initial diagnosis, without being ever submitted to hysterectomy-bilateral salpingo ophorectomy she is alive, asymptomatic, without evidence of progressing disease.

Discussion

Peritoneal keratin granuloma is a rare lesion included under granulomatous lesions of the peritoneum (Clement, 1995). Such peritoneal reaction can be infectious or non-infectious in etiology (Clement, 1995). The non-infectious type can be secondary to neoplasms of female genital tract, like endometrioid adenocarcinoma with squamous differentiation of the endometrium and ovary, atypical polypoid adenomyoma of endometrium and in association with ruptured dermoid cysts.

After revision of literature, only 32 cases were reported to date. At surgery these lesions can be misinterpreted as disseminated carcinomas (Tripathy et al., 2010). Montes et al. (1961), in 1961, were the first to describe a case of well-differentiated adenocarcinoma of the...
constituting the largest review of cases published in literature. It was the first time that such peritoneal lesions were described to be related to endometrioid adenocarcinoma of the ovary (five cases). In 2006, Wu et al. (2006), described the other case of peritoneal keratin granuloma in association with ovarian adenocarcinoma. It was suggested by Kim and Scully (1990) that tearing of the capsule of the tumor or malignant cell penetrating the ovarian surface was the way of cells in entering the peritoneal cavity. The last two published cases, by Van der Horst and Evans (2008) in 2008, refer to carcinomas of the endometrium also.

Lack of or short follow-up in some cases and post-operative radiotherapy, chemotherapy or both, which might have influenced the natural course of any post-operative residual peritoneal lesions, makes more difficult to interpret the real prognostic significance of these lesions. Some authors suggest that they have no prognostic significance when no viable cells are found in the granulomas.

We report a case with twelve years follow-up of an endometrioid adenocarcinoma of the endometrium with disseminated peritoneal keratin granulomas and viable tumor implants. Interpreted as disseminated disease lead to a palliative approach with only brachytherapy and hormonal therapy, the outstanding survival could suggest no adverse effect on the prognosis of such peritoneal lesions even with viable tumor implants. This might be justified due to immunological factors recognizing the tumor as a threat eliciting a reaction that lead to regression of the tumor as reported in patients with esophageal squamous carcinoma treated with preoperative chemoradiotherapy in which keratin granulomas and ghost cells are described as a feature of disease regression (Kim and Scully, 1990).

The prognostic significance of keratin peritoneal granulomas with or without viable tumor implants is difficult to assess because of the small number of cases in the literature.

It is important to exhaustively sample the peritoneal granulomas seeking for viable cells to further understand the behavior of such peritoneal lesions when associated with malignancy.

Conflict of interest statement
The authors don't have any relationships with companies that may have potential interest in the information contained in the manuscript.

References