

A case of a borderline mucinous tumor of the ovary with sarcoma-like mural nodules producing granulocyte colony-stimulating factor

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Summary

Borderline ovarian tumor with sarcoma-like mural nodule is rare. Malignant mural nodules usually occur in the wall of an atypical proliferative mucinous tumor or a mucinous carcinoma. The authors report one case of unfavorably progressive borderline tumor of the ovary with sarcoma-like mural nodule that produced granulocyte colony-stimulating factor (G-CSF).

Key words: Borderline ovarian tumor; Mural nodule; Granulocyte colony stimulating factor.

Introduction

Mucinous ovarian tumors with sarcoma-like mural nodules are rare [1]. Among diverse histologic features of mural nodules, sarcoma-like (benign) and malignant nodules are generally categorized. It has been reported that borderline tumors with sarcoma-like mural nodules have more favorable outcomes than malignant nodules [1]. Leukemoid reactions (white blood cell [WBC] count $> 50 \times 10^9/l$) and production of granulocyte colony-stimulating factor (G-CSF) are observed in many types of tumors, but seldom reported in gynecologic tumors.

The authors report a rare case of a progressive borderline mucinous tumor of the ovary with sarcoma-like mural nodules producing G-CSF.

Case Report

A 33-year-old woman presented to the Clinic for evaluation of abdominal fullness. A 22 cm by 10 cm multi-loculated cystic mass was noted on pelvic computed tomography (CT), suggesting a mucinous cystic adenoma or adenocarcinoma (Figure 1A). The levels of CA-125 and CA 19-9 were 1,516 and 27.21 U/ml, respectively. On exploratory laparotomy, a 18 cm by 13 cm ruptured cystic mass was noted on the left ovary. The cyst contained multiple chambers, solid portions, and areas of papillary growth (Figure 1B). The final diagnosis was a borderline mucinous tumor of the ovary with sarcoma-like mural nodules.

Along with nodularities on liver surface, cancerous masses had spread to the omentum and to small and large bowel mesentery. Staging surgery, including a total hysterectomy, bilateral salpingo-oophorectomy, total omentectomy, partial peritonectomy, and pelvic and para-aortic lymphadenectomies, were performed. Poorly-differentiated malignant tumor involved the ascites, omentum, bowel mesentery, and 22 of 86 lymph nodes. Immunohistochemistry staining was strongly positive for vimentin, actin, CK7, and CK20, and negative for desmin, S-100, and CEA. Severe leukocytosis (WBC count, $53.6 \times 10^9/l$) was noted and expression of G-CSF was strongly positive on

the borderline tumor and the sarcoma-like nodules based on immunohistochemistry (Figure 1C).

Two small nodules were noted in the right middle and lower lobe fissures of the lungs, suggesting metastasis, and rapidly progressing new masses were demonstrated in the peritoneal cavity, suggestive of carcinomatosis peritonei on a post-operative 12-day CT scan (Figure 1D). An incisional wound hernia developed, for which she underwent a primary wound closure on the 13th post-operative day. She passed away without recovery from disease in hospital on day 22.

In this report, the intense neutrophilic infiltration of the tumor and the extremely high G-CSF concentration suggested that such a phenomenon might be involved in dramatic tumor growth.

Discussion

Pratt and Cully first reported sarcoma-like mural nodules in mucinous ovarian tumors in 1979 [1]. In their report, none of the cases had spread to the ovary and the type of mural nodules had no effect on prognosis. Pratt and Scully suggested dividing mural nodules into either benign or malignant types [2]. Compared to malignant nodules, which are liable to recur and be fatal in 50% of cases, sarcoma-like nodules are associated with a favorable outcome [3].

A leukemoid reaction is diagnosed when a WBC count $> 50 \times 10^9/l$ without bone marrow involvement is shown and is observed in association with the production of G-CSF in a wide variety of tumors, including carcinomas of the lung, stomach, and pancreas. In proportion to the degree of leukocytosis, G-CSF expression has been implicated in the progression of malignant behavior in non-hematopoietic cancers. Thus a high mortality rate has been reported among patients with leukemoid reactions, but only one report of ovarian cancer associated with a leukemoid reaction exists in the English literature [4].

This is the first case of a borderline mucinous tumor with sarcoma-like nodules associated with an intense paraneoplastic neutrophilic leukemoid reaction related to the production of G-CSF. Recurrences are rare and slow

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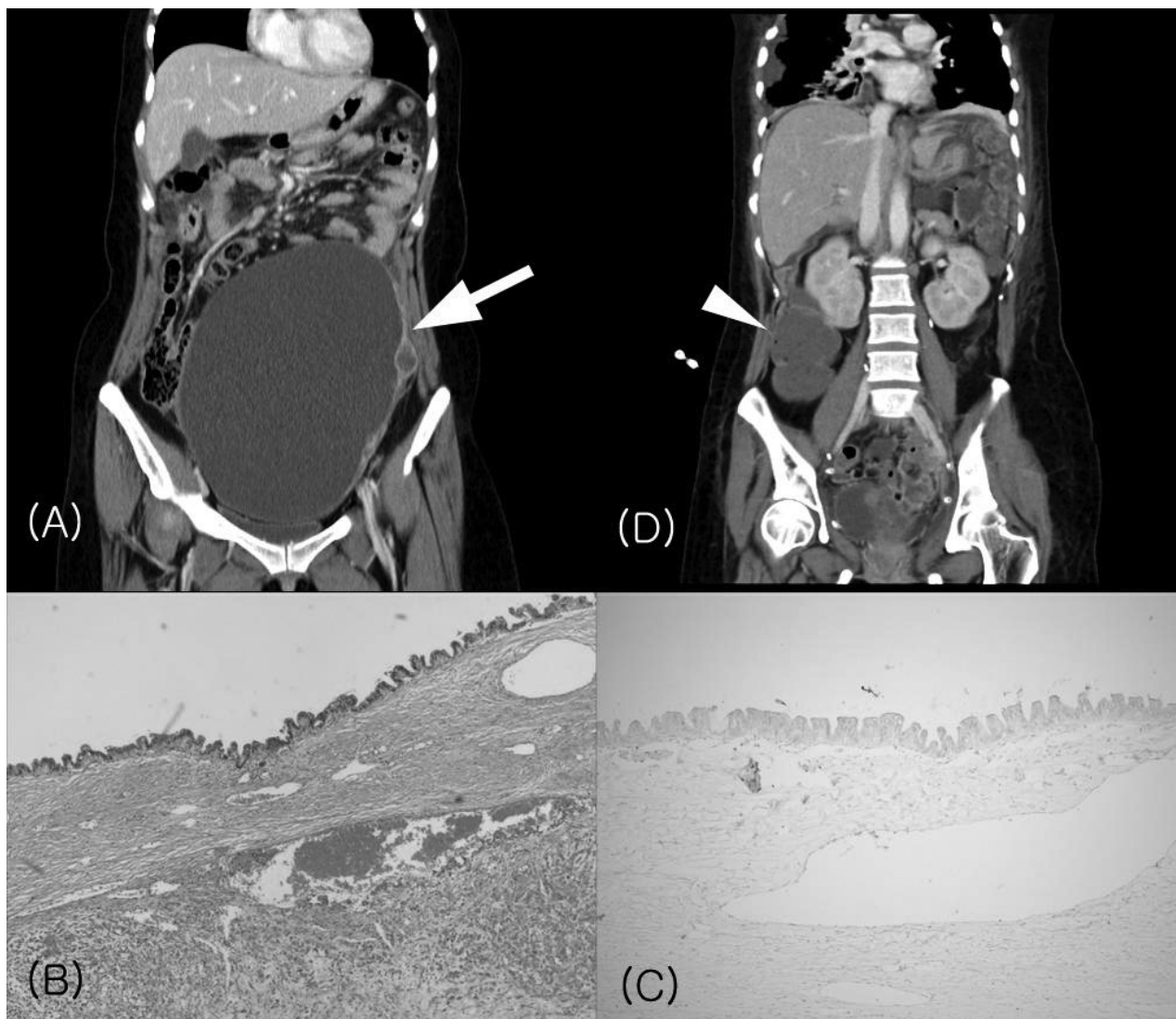


Figure 1. — (A): Large abdominal mass (arrow) in pre-operative CT scan (B): Borderline tumor with sarcoma-like nodules in the ovarian tumor. Pseudostratified mucinous cell lining gland and thickened wall are concurrently shown and atypical sarcoma-like cells are scattered on the nodules. Magnification power X 100. (C): Expression of G-CSF in sarcoma-like nodules. Dot-like staining was scattered throughout the cytoplasm in a “Golgi pattern.” Inflammatory cells with a small nucleus were also stained by G-CSF (1:100 dilution). Magnification power X 100. (D): Post-operative follow-up CT at 12 days showing a newly-developed pelvic mass indicative of carcinomatous peritoneii (arrow head).

to occur through a metastatic nodule in a borderline ovarian tumor. However a highly-elevated G-CSF led to rapid cellular proliferation and this explained the residual and re-growth of tumor cells two weeks after operation. A leukemoid reaction is associated with a poor prognosis in patients with borderline ovarian tumors.

Acknowledgement

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