Adenofibromasarcoma originating from a mural nodule of ovarian serous cystadenoma

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Summary

A 83-year-old woman received bilateral salpingo-oophorectomy and hysterectomy due to a provisional diagnosis of ovarian cystic tumor. The tumor had a unilocular cystic cavity demonstrating serous cystadenoma and a solid mural nodule representing a biphasic pattern with mesenchymal and glandular components. The glandular elements were composed of benign serous cells, whereas the mesenchymal components consisted of an admixture of fibromatous stromal cells without atypia and sarcomatous overgrowth. The area of transition from a fibromatous component to sarcomatous overgrowth was identified. After a 2-year follow-up, there were no signs of tumor recurrence or systemic disease. To the authors' knowledge, this is the first reported case of adenofibrosarcoma originating from a mural nodule of ovarian serous cystadenoma.

Key words: Adenofibromasarcoma mural nodule serous tumor ovary.

Introduction

Although there have been sporadic reports in the literature of a carcinomatous mural nodule in a benign cystic common epithelial tumor [1-3], the occurrence of a sarcomatous mural nodule has been reported only in one case, which was mucinous cystadenoma with a mural nodule of fibrosarcoma [4]. Adenosarcomas differ from carcinosarcomas in that the epithelial component is benign and from ordinary adenofibromas in that the stroma is sarcomatous. We describe the first reported case of adenofibrosarcoma originating from a mural nodule of ovarian serous cystadenoma.

Case Report

An 83-year-old woman, gravida 2, para 2, was admitted to the hospital complaining of backache and lower abdominal pain. Her medical and surgical histories were negative. A pelvic examination confirmed the presence of a large mobile, pelvic-abdominal mass. Preoperative computed tomography revealed a partly solid, partly cystic ovarian tumor measuring 16 x 15 x 10 cm. There was no ascites. Cystoscopy and rectoscopy were negative. The patient's tumor marker profile was as follows: CA 19-9 (56 U/ml; normal < 37 U/ml), CEA (2.4 ng/ml; normal < 2.5 ng/ml), CA-125 (11 U/ml; normal < 35 U/ml). At explorative laparotomy, a large mass originating from the left ovary was found. There was no ascites or obvious lymph node swelling. The right ovary and uterus appeared to be intact. Bilateral salpingo-oophorectomy and hysterectomy were performed. The left ovarian cystic mass measured 15 cm in diameter. The cut surface of the left ovary showed a predominantly unilocular cyst. The cut surface of the left ovary showed a predominantly unilocular cyst. There was a protruding solid mural nodule, measuring 6 x 6 x 4 cm (Figure 1).

Histologically, the glandular elements were composed of benign serous cells. No nuclear atypia were demonstrable within the cystic tumor component. The mural nodule showed a biphasic pattern with mesenchymal and glandular components. The glandular elements were composed of benign serous cells (Figure 2). The mesenchymal components consisted of an admixture of fibromatous stromal cells without atypia and sarcomatous overgrowth. The sarcomatous cells had marked nuclear atypia and a high mitotic index (more than 5/10 high-power fields). Hypercellular bundless of spindle-shaped tumor cells arranged in a herringbone pattern were visible (Figure 3). An area of transition from a fibromatous component to sarcomatous overgrowth was identified (Figure 4). The immediate postoperative course was uneventful. The CA19-9 level analyzed one week after surgery was normal (30 U/ml). Clinical follow-up including abdominal sonography and magnetic resonance imaging has been negative for over two years.

Discussion

A solid mural nodule within a cystic ovarian tumor is extremely rare. Prat and Scully [5] described seven cases of "sarcoma-like nodules" in ovarian mucinous tumors based on light microscopy and clinical information. They concluded that these nodules represented reactive sarcoma-like lesions resulting from hemorrhage in the cyst walls. The epithelial components of the tumors were borderline malignancy and well-differentiated carcinoma. The same authors also reported two cases of true sarcomatous nodules within mucinous ovarian tumors [4]. One of the tumors was a fibrosarcoma associated with a mucinous cystadenoma; the other was an undifferentiated sarcoma in a mucinous cystadenocarcinoma. The present case is the second case of a fibrosarcoma and the first case of an adenofibrosarcoma arising from a mural nodule within a benign ovarian cystic tumor.

The histology of the present case was consistent with adenosarcoma. The glands lined by a variety of benign müllerian epithelia were covered by fibrous tissue. Architectural or cytological atypia was absent. The most problematic lesion in the differential diagnosis of adenosar-

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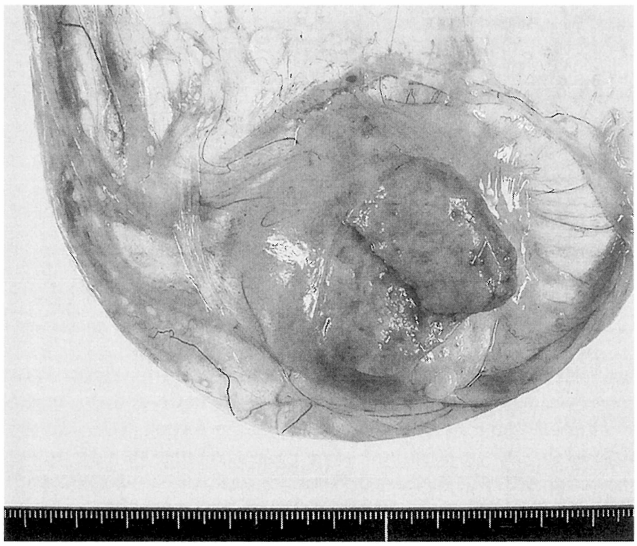
Fig. 1



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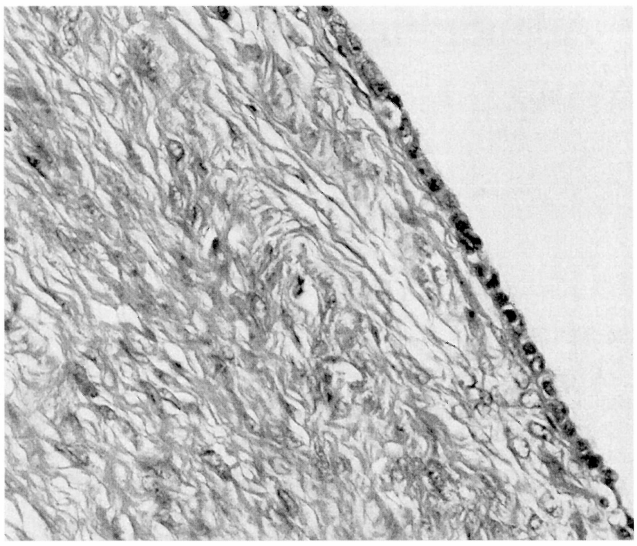
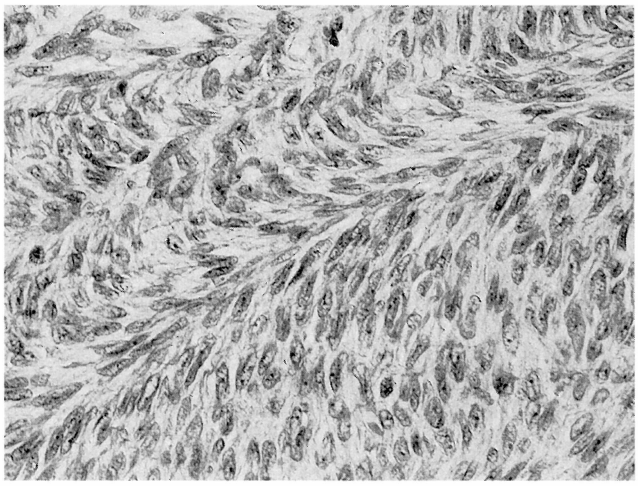


Fig. 3



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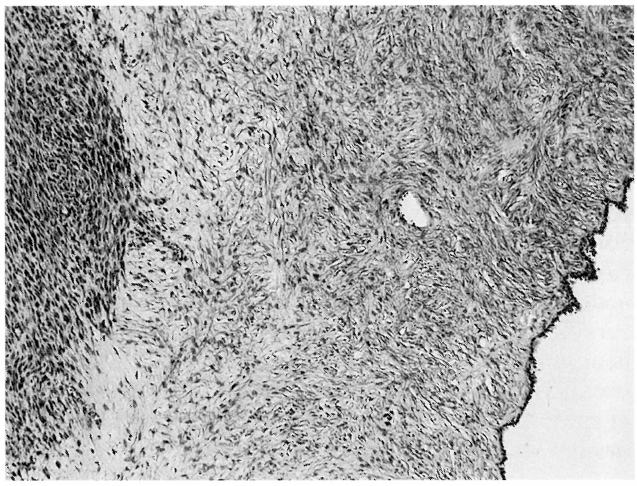

Figure 1. — A solid 6 x 6 x 4 cm nodule in the wall protruding into the lumen of a unilocular cyst.
Figure 2. — A single-layered benign epithelium covered by a dense fibrous component (H&E x 400).
Figure 3. — Fibrosarcoma composed of spindle-shaped tumor cells arranged in a herringbone pattern with easy identifiable mitotic figures (H&E x 400).
Figure 4. — Area of transition from a fibromatous component to sarcomatous overgrowth is seen (H&E x 100).

coma is adenofibroma. Clement and Scully [6] commented that they have diagnosed adenosarcoma in contrast to adenofibroma when one or more of the following are present: 1) a stromal mitotic count of two or more per 10 high-power fields; 2) marked stromal cellularity; 3) significant stromal cell atypia. In our case, the neoplasm was densely cellular, the spindle-shaped cells being arranged in a herringbone pattern. There was a moderate to marked degree of pleomorphism and at least five mitotic figures per 10 high-power fields.

Although little is known about the histogenesis of extrauterine adenosarcoma, there is a general impression that fibrosarcomas represent dedifferentiated counterparts of ovarian fibromas rather than occurring as a malignant tumor de novo [7, 8], and strength is lent to this view by a report of a fibrosarcoma, which developed in an eight-year-old girl with multiple ovarian fibromas as a component of the nevoid basal cell carcinoma syndrome [9]. In the present case, areas of transition from a fibromatous component to sarcomatous overgrowth were identified. These findings support the hypothesis that the sarcomatous component is derived from malignant stromal change of the fibromatous component.

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