Sebaceous Carcinoma Arising in Mature Cystic Teratoma of Ovary

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Mature cystic teratoma (MCT) is the most common ovarian tumor.1 Malignant transformation of a component of a MCT is a very rare event, occurring in less than 2% of cases,2 with squamous cell carcinoma as the most common malignancy.1,3 Cutaneous-type adnexal neoplasms including basal cell carcinoma, melanoma, and apocrine adenocarcinoma have also been reported as associated malignancies with MCT.1,3 Sebaceous carcinoma arising within a MCT has been rarely reported. To our knowledge, there have been only seven prior reports of sebaceous carcinoma arising within a MCT of the ovary.6-12 Here, we present an additional case of a sebaceous carcinoma, along with a review of the previous reports.

CASE REPORT

A 69-year-old gravida 4, para 4 Korean woman visited our hospital with a two week history of pelvic pain. Ultrasonography and computed tomography of the abdomen demonstrated a large pelvic mass measuring 22.0 cm in maximal diameter and with a small amount of ascites. Her preoperative serum cancer antigen 125 level was elevated at 430.5 U/mL. Together, these findings were suggestive of malignancy. A total abdominal hysterectomy with bilateral salpingo-oophorectomy and partial omentectomy were performed. A massive left ovarian mass and omental cake were noted in the pelvic cavity, and numerous nodules (1-2 cm in size) were scattered in the peritoneum and along the intestinal serosal surface. A neoplastic implant measuring 1.5 cm in diameter was also observed at the posterior portion of the uterine body. The right ovary was unremarkable. The resected left ovary measured 22.0 cm in diameter and weighed 2,180 g. The outer, capsular surface of the ruptured left ovary appeared ragged with scattered tumor implants measuring up to 1.2 cm in diameter. On the cut section, the left ovary was replaced by a unilocular cyst filled with keratin-like material and brownish-serous fluid. The luminal surface of the cyst was smooth, but a luminally-protruding and outer expanding mass measuring 6.0 cm in diameter was noted. The cut surface of the mass was relatively grayish-white in color and firm in the luminal portion and was tan-colored and friable with hemorrhagic necrosis in the outer, expanding portion (Fig. 1). Microscopically, the smooth cystic wall was lined by stratified squamous epithelium with underlying sebaceous glands and other skin adnexal structures, findings consistent with a...
typical mature cystic teratoma. Benign squamous epithelium was abruptly replaced by a nodular arrangement of germinative cells with a pushing border, which protruded into the cyst lumen (Fig. 2). The nodular portion showed an alternatively dark and white area, which corresponded to generative cells (dark) and sebaceous cells (light) with cytoplasmic lipid vacuoles. There was no cytologic atypia or sparse mitosis. Taken together, these findings led to a diagnosis of sebaceous adenoma. Beneath the nodular portion, infiltrating trabeculae or nests of atypical cells were noted. The infiltrating portion was mostly separated but was focally contiguous with the sebaceous adenoma (Fig. 3A). Infiltrating cells exhibited conspicuous vacuoles in the cytoplasm and remarkable nuclear pleomorphism, prominent nucleoli, and frequent abnormal mitoses (Fig. 3B). There was no peripheral nuclear palisading or cleft-like spaces between the lobules. Tumor cells were immunohistochemically positive for cytokeratin (CK) 7 (Fig. 4A), CK19, high molecular weight CK, epithelial membrane antigen (EMA) (Fig. 4B), carcinoembryonic antigen (CEA) (Fig. 4C), and p63 (Fig. 4D), but stains were negative for CK20, p53, vimentin, human placental alkaline phosphatase, α-inhibin, S100 protein, c-erbB-2, estrogen receptor, and progesterone receptor. Periodic acid-Schiff (PAS) was negative in the large, cytoplasmic inclusion. The uterine myometrium showed direct infiltration of the sebaceous carci-
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The patient developed sepsis two weeks after diagnosis and was discharged home with palliative care.

DISCUSSION

As the most common ovarian germ cell tumor, MCT undergoes malignant transformation in less than 2% of cases. MCTs with malignant change are usually larger than conventional MCT and have a grossly cauliflower-like appearance or a mural nodule protruding into the cyst lumen. Clinical presentation of malignant MCT depends on the extension and histological type of the secondary malignancy. Squamous cell carcinoma serves as the most common secondary tumor seen in 80% of malignantly-transformed MCT cases. Other malignancies have also been associated with MCT including adenocarcinoma, adenosquamous carcinoma, sarcoma, and melanoma. Normal skin adnexal structures, including sebaceous glands, are frequently observed in MCTs, but malignant, neoplastic transformation in such tissues is rare. Sebaceous cell carcinoma in association with MCT has been infrequently reported with only seven cases published.

Sebaceous carcinomas arising within MCTs have similar gross and microscopic findings with their cutaneous counterparts. However, tumor cells are frequently large and can have a squamoid appearance with occasional squamous pearls. In such cases, sebaceous carcinoma must be distinguished from squamous carcinoma with hydropic change. Occasionally, malignant squamous cells may accumulate glycogen with clear cytoplasm. This can be confused with large, foamy sebaceous cells. The present case also revealed squamoid differentiation. In contrast, the large, foamy sebaceous cells were PAS negative, a finding that confirmed the absence of glycogen, and the majority of tumor cell nests had sebaceous differentiation. Basal cell carcinoma was also considered in the differential diagnosis of sebaceous carcinoma with basaloid differentiation and inconspicuous li-
though our patient has been lost to follow-up, her general condition was poor with associated secondary sepsis. Her prognosis was therefore deemed poor.

In summary, cases of sebaceous carcinoma arising within an ovarian MCT are extremely rare. The case presented here appears to be the first to report both a sebaceous carcinoma arising within an ovarian MCT combined with a sebaceous adenoma. The behavior of a sebaceous carcinoma arising from an ovarian MCT remains poorly understood due to their rarity. The present case was associated with metastasis and peritoneal seeding whereas previous reports mostly had uneventful outcomes.

Conflicts of Interest
No potential conflict of interest relevant to this article was reported.

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