Combined Microcystic Adnexal Carcinoma and Squamous Cell Carcinoma Arising in the Ovarian Cystic Teratoma —A Brief Case Report—

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Malignant transformation of mature cystic teratoma of the ovary can develop with an incidence of 1-2%. Squamous cell carcinoma is the most common malignant tumor arising in benign cystic teratoma. The authors have recently experienced a case of combined microcystic adnexal carcinoma and squamous cell carcinoma arising in a benign cystic teratoma of the ovary in a 72-year-old Korean woman. The right ovarian cystic mass had been ruptured and firmly adhered with salpinx and omental fat tissue on operation. Thickened cystic wall with yellowish white solid infiltrative lesion was noted grossly, and two different malignant tumors of microcystic adnexal carcinoma exhibiting both eccrine and hair follicular differentiation and squamous cell carcinoma were observed microscopically. PAS and CEA positivities suggested eccrine differentiation in areas of microcystic adnexal carcinoma.

Key Words : Microcystic adnexal carcinoma, Squamous cell carcinoma, Cystic teratoma, Ovary.

INTRODUCTION

Mature cystic teratoma of the ovary is common in woman of child-bearing age. In 1-2% of mature teratomas, malignant transformation can occur (Kelly and Scully, 1961). The most common malignant tumor arising in mature teratoma is squamous cell carcinoma which accounts for 80-90%, and the remainder consist of adenocarcinoma, sarcoma, undifferentiated carcinoma and papillary carcinoma (Stamp, 1983). Recently the authors have experienced a case of combined microcystic adnexal carcinoma of the sweat gland and squamous cell carcinoma arising in mature cystic teratoma of a 72-year-old woman.

CASE REPORT

A 72-year-old woman complained of persistent lower abdominal pain for several days. On physical examination, tenderness was noted in the lower abdomen. Her obstetric history was para 6 and 6 liveborns, and her menarche was 19 years of age, and menopause, 46 years of age. Family history and past history were not contributory. Complete blood count revealed leukocytosis ($13,800/\text{mm}^3$), and tumor markers were within normal ranges; Alpha-fetoprotein 2.19 ng/ml, carcinoembryonic antigen (CEA) 1.3 mg/ml, CA 125 0.20 u/ml, human chorionic gonadotropin (HCG) 0.85 mIU/ml. On operation, the right ovary was found to be ruptured previously, and a right salpingo-oophorectomy and an incidental appendectomy were performed.

Pathologic findings: The right ovarian cystic mass was 11.0×7.0×5.0cm in dimension, and the outer surface was partly covered with necrotic material

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Fig. 1. Cystic wall shows thickened solid infiltrative lesion extending to adhered salpinx and omentum.

Fig. 2. Linings of thickened cystic wall are focally of malignant squamous cells with downward infiltrative growth.

and adherent to the omental fat tissue. The right tube was not discernible externally. The unilocular lumen was filled with dark reddish brown greasy or mucoid materials, and the cystic wall was focally thickened forming a solid yellowish to grayish white, firm and fibrotic mass with focal necrosis measuring 4X3cm across. (Fig. 1) This infiltrative lesion was extending to the adhered salpinx and omental adipose tissue. Microscopically thin cystic wall was lined by stratified squamous or single or pseudostriated cuboidal or mucin-laden columnar cells. The lining epithelium of the solid area was focally sloughed or composed of flattened cuboidal cells and mucin-containing columnar cells. Foci of malignant keratinizing squamous epithelium with infiltrative growth into the stroma, which was typical for squamous cell carcinoma, were seen. (Fig. 2) In areas, neoplastic tubules, cords, and nests were scattered in the fibrohyalinized stroma, and tadpole-

shaped tubular structures were occasionally found. (Fig. 3) These tubular structures were surrounded by unevenly stratified polygonal cells with modest nuclear pleomorphism and infrequent mitosis and partly filled with acidophilic secretory materials, which were positive for PAS stain and for CEA on immunohistochemical stain. CEA positivity was seen along the luminal borders and faintly in the cytoplasm. Just beside this tubule-forming tumor, there were atypical squamous cell clusters or horny cysts considered as hair follicular differentiation. (Fig. 4)

DISCUSSION

Malignant transformation of mature teratoma is more common over the fifth decade in contrast to mature cystic teratoma which is common in the second or third decades. Clinically most patients complain of non-specific symptoms of intraabdomin-

Fig. 3. Variable tubular structures with tail-like extension are found in the dense collagenous stroma.

Fig. 4. Squamoid cell clusters and horn cysts suggesting hair follicular differentiation are present in areas.

al mass and persistent lower abdominal pain, but sometimes rapidly enlarged mass has been noticed (Stamp, 1983). Gross findings suggesting malignant change are adhesion of pelvic organs, presence of thickened cystic wall or nodular lesion within the teratoma besides Rokitansky protuberance, and hemorrhagic necrosis (Kelly and Scully, 1961). Any kind of malignant tumor can arise from mature teratoma but mostly malignant tumors show single histologic type. Some malignant tumors arising in mature teratoma of the ovary had been previously ruptured on operation due to necrosis and marked adhesion with adjacent organs, and hematogenous or lymphatic spread was accompanied in 64% of the cases (Peterson, 1957). Prognosis depends on clinical stage, histologic type and degree of differentiation of the tumor (Kelly and Scully, 1961), but Stamp(1983) reported no relationship between prognosis and histologic type of the tumor, and

mean survival time was 8-23 months. Kelly and Scully (1961) reported that 5-year survival rate was 15% in ruptured cases compared to 63% in non-ruptured cases.

In the presenting case, two different histologic types of squamous cell carcinoma and microcystic adnexal carcinoma were noted. Squamous cell carcinoma is a well-known malignant tumor arising in mature teratoma, but microcystic adnexal carcinoma arising in mature teratoma has not been reported in the English literature. Microcystic adnexal carcinoma of the skin is a rare malignant tumor described by Goldstein et al. in 1982. This tumor is slow growing and locally infiltrating, and shows both eccrine and hair follicular differentiation, thus histologically suggesting pluripotent adnexal keratinocytic origin (Goldstein et al., 1982; Cooper et al., 1985). Characteristic microscopic findings of microcystic adnexal carcinoma of the skin are squamous cell

clusters, horny cysts and various tubular structures including tadpole-shaped ducts, sclerosing fibrous stroma, and infiltrative growth in the perineural and subcutaneous fat tissue (Goldstein et al., 1982; Cooper et al., 1985). Ductal or acinar cells of eccrine or apocrine differentiation reveal positive reaction for CEA on immunohistochemical study but the tumor cells of hair follicular differentiation are not reactive for CEA (Nickoloff et al., 1986), and CEA positivity was noted in the area of eccrine differentiation in the presenting case.

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