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ADENOID SQUAMOUS CELL CARCINOMA ARISING IN A BENIGN CYSTIC TERATOMA OF THE OVARY

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Synopsis A rare case of adenoid squamous cell carcinoma arising in a benign cystic teratoma was reported in a 51-year-old woman. The tumor was basically a keratinizing type of squamous cell carcinoma, but had the focal adenoid structures with dyskeratotic cells inside the lumen. The prognosis was as bad as in the usual squamous cell carcinoma of the ovary, and the patient died in spite of a combination chemotherapy of bleomycin and mitomycin C.

Key words: Adenoid squamous cell carcinoma • Ovary

Introduction

Adenoid squamous cell carcinoma is an uncommon variant of the squamous cell carcinoma and was originally described in the skin[6]. In gynecology, it was reported in the vulva[5)10]. In this paper, we describe a rare adenoid squamous cell carcinoma arising in a benign cystic teratoma of the ovary.

Case Report

A 51-year-old gravida 7, para 5 woman was admitted to Osaka University Hospital on September 21, 1977 because of a rapidly growing lower abdominal tumor with a dull pain and a remarkable emarciation. Her past history revealed that a tumor of hen egg in size was palpable in the right adnexal region in 1966 when D & C were performed, but left untreated. Her menarche was at age 18. Since then, she had had a regular menstruation every 30 days.

Physical examination revealed a hard tumor of over-adult head in size in the lower abdomen. Her stature was 159 cm and her body weight 49 kg. Her blood pressure at admission was 110/70 mmHg. No metastatic tumor was palpated in the inguinal, axillary and Virchow lymph nodes.

Laboratory examination revealed normal values in the peripheral blood analysis, the serum electrolytes, and the liver functions. The findings obtained by hysterosalpingography and ultrasonic echograms were in favor to the diagnosis of a solid tumor of the right ovary.

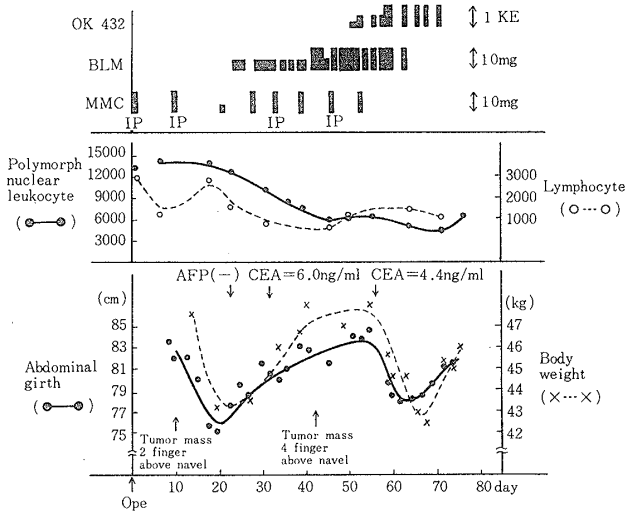
Laparotomy was performed on September 27, 1977. About 50 ml of slightly bloody ascites was in the peritoneal cavity. A 10×10 cm tumor arising in the right ovary was predominantly solid, but partly cystic and adhered to the surrounding tissues such as peritoneum, intestine and urinary bladder. Behind the tumor, the uterus and the left ovary were tightly adhered to the Douglas cavity. Many small metastatic foci were also found in the intestinal wall. Only the major tumor of the right ovary was resected and a dose of 10 mg of mitomycin C was administered intraperitoneally.

After surgery, the patient was treated by a combination chemotherapy of bleomycin and mitomycin C, and an immunotherapy of OK-132, as shown in Fig. 1. The patient died, however, of a hypovolemic shock caused by ileus 80 days after surgery.

Pathologic Findings

Gross. The tumor was of an adult head in size and cystic filled with greasy substance and long hairs. Part of the wall was, however, occupied by the solid tumor mass which proliferated predominantly to the peritoneal

Fig. 1. Post-operative treatments and clinical course.



cavity but partly into the lumen of the cyst (Photo. 1).

Microscopic. The cystic part of the tumor was composed mainly of the fibrous tissue with hairs and a few small round malignant cells covering the surface (Photo. 2). The solid part was basically a squamous cell carcinoma showing various histological patterns. The areas projecting into the lumen of the cyst and the peritoneal cavity were papillary and lined by one to multiple layers of small round immature squamous carcinoma cells (Photo. 3). Also, the cornification and keratinization with pearl formations were observed in other places (Photos. 4 and 5). The characteristic histology of the present tumor, however, was a focal formation of adenoid structures and the presence of dyskeratotic cells inside the lumen (Photo. 6).

Discussion

Malignant change of benign cystic teratoma of the ovary was relatively rare and extensively reviewed by Peterson[8] in 1957, by Climie and Heath[1] in 1968, and recently by Krumerman and Chung[4] in 1977. According to these reports, most of the malignancies are squamous cell carcinoma. Adenocarcinoma and carcinoid are much less, and sarcoma is extremely

Photo. 1. Macroscopic appearance of the tumor, showing the solid mass arising in the wall of a benign cystic teratoma.

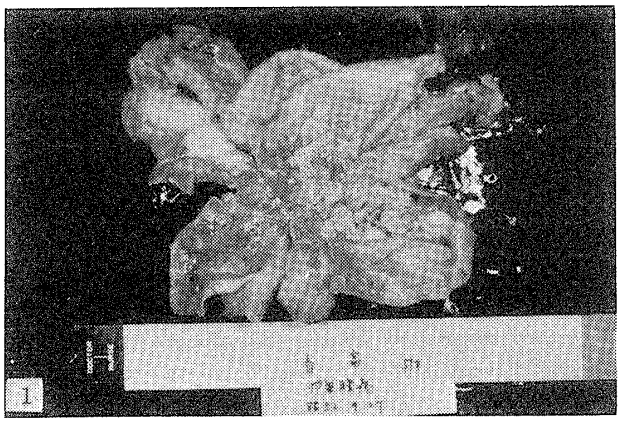


Photo. 2. Fibrous tissue with hairs and a few small malignant cells, composing of the cystic wall of the tumor. Hematoxylin-Eosin, ×100

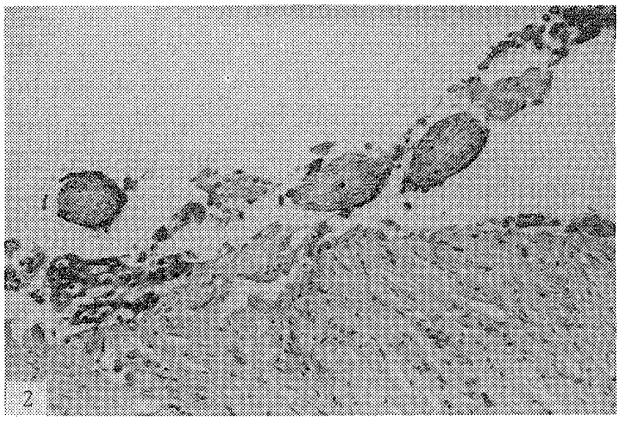


Photo. 3. Papillary growths of immature squamous carcinoma cells, projecting into the peritoneal cavity. Hematoxylin-Eosin, ×100

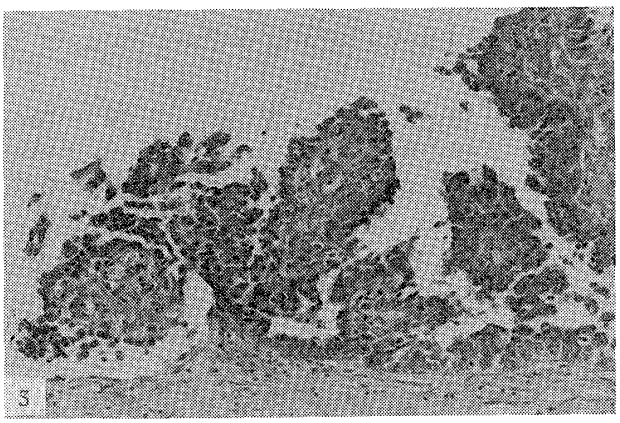


Photo. 4. Cornification of squamous cell carcinoma, seen in a few places. Hematoxylin-Eosin, ×100

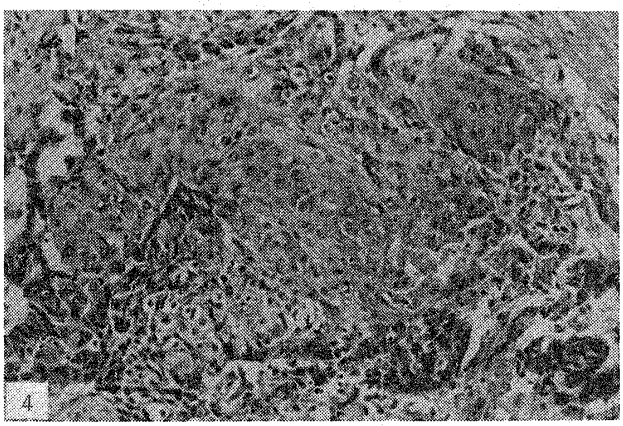


Photo. 5. Pearl formation of squamous cell carcinoma, seen in some places. Hematoxylin-Eosin, ×100

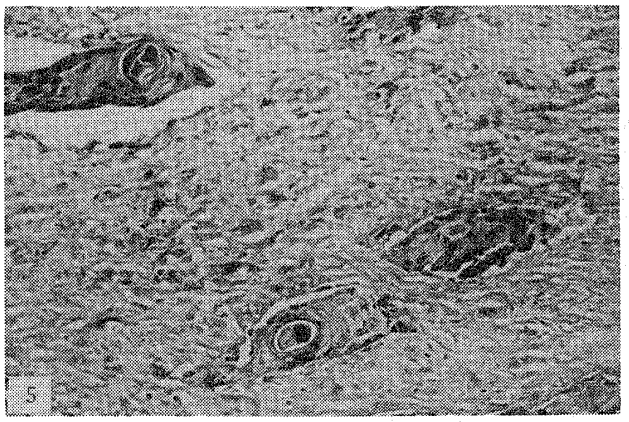
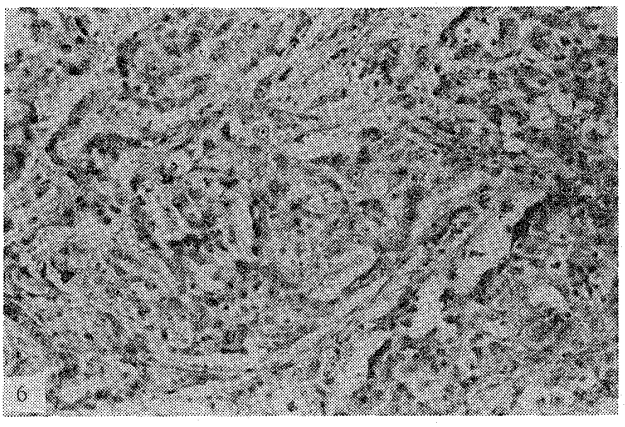


Photo. 6. Focal adenoid structures and dyskeratotic cells inside the lumen, characteristic of the present tumor. Hematoxylin-Eosin, ×100



rare. In Japanese literature reviewed by us[2] in 1977, 38 squamous cell carcinomas, 9 adenocarcinomas, 1 carcinoid and 1 sarcoma were reported as arising in a benign cystic teratoma. Adenoid squamous cell carcinoma, however, was never described in a benign cystic teratoma of the ovary.

Adenoid squamous cell carcinoma was first described in the sun-exposed areas of the skin by Leber[6] in 1947, and considered to have a causative relationship to sun light. Recently, however, this has been reported to occur in the vulva[5][10] and the oral mucosa[9]. The tumor was characterized by three histologic criteria: that is, the keratinizing type of squamous cell carcinoma, the adenoid structures lined by squamous cells primarily of one cell thickness, and the dyskeratotic, acantholytic cells in the lumen. This usually occurs as a focal change and rarely in the extensive areas[9]. The present case fulfils these three histological criteria, although they are seen in only a focal area. The pathogenesis of this tumor still remains debatable, but the facts that metastatic lesions may not contain the similar adenoid pattern suggest the local effects for the formation of this characteristic histology[9].

The prognosis was reported to be better in the adenoid squamous cell carcinoma than in the usual squamous cell carcinoma by some[3][11], but not to be better or even worse by others[5][10]. The prognosis of the present case was actually bad and the patient died in spite of a combination chemotherapy of bleomycin and mitomycin C which was reported very effective to the advanced squamous cell carcinoma of the uterine cervix[7].

Acknowledgements

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概要 51歳の7回経妊，5回経産婦の右卵巣から発生した卵巣皮様嚢腫に見られた adenoid squamous cell carcinoma の稀な1症例を報告した．本腫瘍は基本的に keratinizing type の squamous cell carcinoma であるが，部分的に adenoid structures を示し，腔内に dyskeratotic cells を含む特徴的な組織像を示していた．本症例の進行度は FIGO 分類の stage Ⅲ に相当し，小骨盤腔を超えて小腸その他腹膜内に広汎に拡がつていたため，右卵巣の主腫瘍切除が行われたのみである．術後，婦人科扁平上皮癌に有効とされる B-M 療法が試みられたが，予後不良で術後80日目に死亡した．