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Squamous Cell Carcinoma Arising in Benign Cystic Teratoma of the Ovary; Report of Two Cases

Gyung Byung Roh, Sung Ho Lee, and Soon Eung Kim

Departments of Gynecology and Obstetrics, and Pathology
Yonsei University College of Medicine

ABSTRACT

Two cases of squamous cell carcinoma arising in benign cystic teratoma of the ovary are reported because of the rarity of this condition. No similar cases have been recorded yet in Korean literature. Furthermore, two different co-existing malignant tumors were encountered in one of these cases; a squamous cell carcinoma arising in a benign cystic teratoma of the right ovary and a serous cystadenocarcinoma of the opposite ovary. The latter metastasized to the right ovary. Important points in diagnosis and management of this complication in the operating room are briefly discussed.

Benign cystic teratoma, the so-called dermoid cyst, is a relatively common ovarian neoplasm. Approximately 11 per cent of all neoplasms of the ovary are of this kind (Peterson et al., 1955). In marked contrast, malignant degeneration of a benign cystic teratoma is rare. Peterson (1957) was able to collect only 222 acceptable cases of malignant degeneration, after an extensive review of every known report on this subject. In his article, the incidence, age, location and size, symptoms, types of malignancy, mode of spread, prognosis and treatment have been discussed in detail.

The following two cases are reported because of the rarity of this condition and to stimulate others to report their cases. As far as the authors know, no similar cases have been reported in Korean literature.

REPORT OF CASES

Case 1. Mrs. H.S.S. (Hospital No. 9921). A 63-year-old, nulliparous Korean woman was admitted to Severance Hospital on February 28, 1959, because of a gradually growing lower abdominal mass, slight fever with chilly sensation, indigestion and anorexia of one month's duration. The menopause was at the age of 48, and no history of abnormal vaginal discharge or bleeding was noted since. The past history was noncontributory except for cholelithiasis at the age of 30 and a cerebral vascular accident one year before admission.

On admission, she was poorly nourished and appeared chronically ill. The temperature was 38°C, the blood prssesure 120/60. Physical examination revealed that both lungs were slightly emphysematous. The heart was normal to auscultation. A round, smooth, somewhat fixed mass measuring 10×10×12 cm was palpable in the lower abdomen. Slight tenderness was noted all over the lower abdomen without rebound tenderness. Pelvic examination revealed the uterus to be retroverted, flexed and atrophic. The cervix was also atrophic, but was normal in appearance. There was a cystic mass of a 4-months' gestation size in the right adnexal region. The mass was slightly tender and partly fixed.

The laboratory findings were as follows: Hb. 10.5 gm%; WBC 1175/mm³, segmented neutrophils 71%, lymphocytes 22%, eosinophils 1%, monocytes 6%, the RBC sedimentation rate 41 mm/hr. (corrected).

Catheterized urinalysis: specific gravity 1029, protein 1+, occasional hyaline and granular casts. Fasting blood sugar was 106 mg %. Non-protein nitrogen 23.2 mg %. Repeated blood cultures showed no growth of organisms. Electrocardiography and cystoscopy were within normal limits. A barium enema was normal execpt for the extrinsic pelvic mass.

On the llth hospital day, laparotomy was performed under nitrous oxide and ether anesthesia. On opening the abdomen, a large reddish-gray cystic mass arising from the right ovary was found. The mass measured about 15 cm in diameter and was rather firmly adherent to the bladder, rectum and right pelvic wall. Most of the tumor was shelled out by blunt and sharp dissection except for an area in which the cyst wall was markely adherent to the right pelvic wall. At this adherent area, the cyst wall was inadvertently ruptured, and a moderate amount of sebaceous material exuded during removal of the cyst and the right tube. The left adnexa and uterus were also removed, althouth the malignant nature of the tumor was not suspected during the operation. The patient's postoperative course was smooth and she was discharged in fair condition. Her general condition was poor, so it was felt she could not tolerate radiation therapy. The patient expired about 4 months after operation with no follow up. No autopsy was granted.

Pathological Findings (S-59-278)

Gross Description: The specimen was submitted in two parts. One part consisted of the right ovary and fallopian-tube, the other of the uterus and left ovary and fallopian-tube. The right ovary was spherical, cystic and unilocular. It measured 11 cm in its greatest diameter. The external surface was grey and red and was focally covered with fibrous adhesions. The cystic wall varied from 1 mm to 5 mm in thickness and contained aggregates of small loculi at some areas. The inner surface of the cyst was grayish-white, fairly smooth and trabeculated at some areas. The cyst was filled with a grayish-yellow, viscid liquid containing greasy sebum, debris of fine hair and several ball-like masses of hair. At one area of the cyst wall, there was a firm, discoid white mass protruding inward and outward, and measuring 4 cm across and 1.5 cm in thickness. On cut section, this mass was white, opaque and contained numerous yellow speckles. A glovoid polypoid mass measuring 2 cm across was attached to one area of the inner surface of the cyst. On cut section, it consisted of yellow fat tissue covered with thin skin with many fine hairs and covered with white opaque greasy sebum. The right fallopian-tube adherent to the cyst was unremarkable. The uterus was atrophic and meaured 7 cm in length including a cervix, 2.5 cm in thickness and 2.5 cm in width. The external os and cervical canal were unremarkable. The myometrium was grayish white, firm and measured 1.2 cm in thickness. The endometrium was grayish-pink in color and measured 0.2 cm in thickness. The left ovary and fallopian tube were unremarkable.

Microscopic Description: The cyst wall was lined with attenuated keratinizing stratified squamous epithelium and contained many sebaceous glands and follicles distended with keratotic plugs in the dermis. No endodermal or mesodermal components were found in the cyst wall. The sections taken from the discoid mass of the cyst wall described grossly revealed nests, cords and sheets of closely-packed polyhedral cells having round or ovoid generally hyperchromatic nuclei varying markedly in size and in chromatin content, abundunt clear or lightly eosinophilic cytoplasm and distinct cell borders. These nests, cords and sheets of tumor cells were separated by thin or broad septa of dense connective tissue focally infiltrated with lymphocytes. There were occasional mitoses, and occasionally cells contained more than one nucleus. No prickles were seen between the cells, but keratin pearls and individual cell keratinization were frequently found. The tumor was necrotic in many areas. It infiltrated throughout the cyst wall and extended to both inner and outer surfaces.

Pathological Diagnoses: Squamous cell carcinoma, moderately well-differantiated, arising in a benign cystic teratoma (dermoid cyst) of the ovary, right. Senile atrophy of uterus showing postmenopausal endometrium. Chronic cervicities.

Case 2 . Mrs. B.S.P. (Hospital No. 10300). A 58-year-old, para VII, gravida VIII, Korean woman was admitted to the gynecological service of Severance

Hospital on April 1, 1959, with the chief complaint of a large palpable abdominal mass of 6 months' duration and slight lower abdominal pain of 2 months' duration. Personal, social and medical history was non-contributory. All pregnancies had been normal, and she had a normal menopause at the age of 40; there had been no subsequent history of bleeding.

On admission, the patient was poorly nourished and moderately prostrated, but not in acute distress. The temperature was 37.8°C, blood pressure 110/80, and respiration 22/min. The physical examination revealed the head, eye, ear, nose and throat to be within normal limits. The cardiorespiratory system was normal. Significant findings were limited to the abdominal and pelvic examination. On abdominal examination, a well-circumscribed cystic mass of 7 months' gestational size occupied the mid-abdominal area. The mass was partly mobile, but no real fluid wave was found. The bowel sounds were normal. The pelvic examination revealed a uterus which was retroverted, atrophic, fairly mobile and non-tender. The abdominal mass did not descend into the pelvis and the adnexal areas were essentially negative.

The laboratory findings were essentially normal except for 3+glucose in the urine. The fasting blood sugar was 215 mg%, and serum amylase 32 Bodansky units. The upper G-I series and barium enema were normal except for the extrinsic mass. Intravenous pyelography showed bilateral hydronephrosis.

After admission to the hospital a surgical consultation was obtained, and it was felt that the patient probably had a pseudopancreatic cyst or mesenteric cyst. Therefore she was transferred to the surgical service. After her diabetes had been controlled, laparotomy was performed 16 days after admission.

At laparotomy, a long pedunculated, smooth-walled and non adherent cyst arising from the right ovary was found. The mass measured about 20 cm in diameter. Also about 20 cc of yellowish peritoneal fluid was present. A right salpingo-oophorectomy was performed. At this point the gynecological service was notified and resumed the care of the patient. Also found at surgery were whitish carcinomatous nodules varying in size from 0.5 mm to 5 cm on the serosal surface of the large intestine, omentum and bladder. The left ovary was enlarged 2 to 3 times normal size and was adherent to the surrounding pelvic wall, the posterior leaf of the broad ligament and an area of the left sacrouterine ligament. In addition to being enlarged, the left ovary was friable, partly necrotic and was of a gritty consistency. The uterus was unremarkable. With the diagnosis of carcinomatosis secondary to a left ovarian adnocarcinoma, a total hysterectomy and left salpingo-oophorectomy were performed. In addition, the metastatic tumors of the bladder and omentum were removed.

The postoperative course was uneventful and the patient was discharged on the 25 th postoperative day with the advice to receive external radiation therapy.

Pathological Findings (S-59-515)

Gross description: Both ovaries and fallopian tubes, the uterus, a mass removed from the bladder, and the greater omentum were submitted for pathological examination. The right ovary was cystic, unilocular and measured 21×20×18 cm. The external surface was of a grayish-white color and was relatively smooth. On opening, the cyst was filled with a yellow turbid fluid containing yellowish-white greasy material and debris of fine hair. The inner surface was gray, interspersed with yellow, greasy, plaque-like areas, smooth at some areas and irregular at other areas. At one area was a grayish-white and yellow, friable plaque measuring 5×3 cm in size and 1.2 cm in thickness. At one corner of the cyst was a segment of fallopian tube including fimbria measuring 7 cm in length and 0.8 cm in diameter, which was unremarkable. The left ovary, adherent to the lateral wall of the uterus, measured approximately 3×3×2 cm. The external surface, covered with fibrous adhesions focally, revealed brown, friable, papillary growths. On cut section, it was partly replaced by a grayish-white solid tumor containing much brown sandy material. The left fallopian tube was adherent to the ovary, measured 7 cm in length and 0.7 cm in diameter and was unremarkable. The uterus was atrophic and measured 9 cm in length including the cervix, 4 cm in width, and 3 cm in height. The peritoneal surface was focally covered with shaggy fibrous

adhesions. The myometrium, measuring 1.5 cm in thickness, was grayish-white, firm and contained many prominently thickened blood vessels. The external os and portio were unremarkable. A piece of greater omentum contained a firm mass measuring 4×3×2 cm. It cuts with an increased resistance, and the cut surface was grayish-white and contained much translucent sandy material.

Microscopic Description: The cyst wall of the right ovary was lined with attenuated, keratinizing stratified squmaous epithelium and contained sparsely scattered atrophic hair follicles and sebaceous glands. At some areas, the inner portion of the cyst wall, including the inner surface, was widely replaced by a tumor composed of fairly closely arranged nests, cords and masses of polyhedral or spindle-shaped cells varying in size. The nuclei were round or ovoid, varied markedly in size and in chromatin content, and contained distinct nucleoli. There were occasional mitoses, and occasionally cells contained more than one bizarre nucleus. The cytoplasm was abundant and eosinophilic. The cell borders were fairly distinct. There was frequently individual cell keratinization, but no definite prickles were found. This tumor was necrotic in many areas. In one area, the external portion of the cyst wall, including the external surface, was replaced by a tumor composed of cysts containing branching papillary cores lined mostly by a single layer of columnar or cuboidal cells with centrally located, round, ovoid or fusiform, generally hyperchromatic nuclei. There were many psammoma bodies varying in size in the fibrous cores of the papillary structures. The left ovary was replaced by a tumor showing histological findings identical with those described just above. The masses found in the greater omentum and bladder proved to be a tumor histologically identical with that of the left ovary. There were similar tumors also on the peritoneal surface of the uterus. There were scattered foci composed of central round or ovoid glands surrounded by tissue resembling endometrial stroma int he myometrim.

Pathological Diagnoses: Squamous cell carcinoma, moderately well-differentiated, arising in a benign cystic teratoma (dermoid cyst) of the right ovary. Papillary serous cystadenocarcinoma of the left ovary, with metastasis to the right ovary, bladder and omentum and extension to the peritoneal surface of the uterus. Adenomyosis of the uterus. Chronic cervicitis. Nabothian cysts of the cervix.

COMMENT

Malignant degeneration occurred in two cases among 15 cases of benign cystic teratoma of the ovary seen at Severance Hospital from September 1958 to August 1961, thus making a far higher incidence (13%) than those generally accepted (1—1.8%) (Peterson, 1957; Abrams et al., 1956). During the same period of time, 32 cases of ovarian malignancy were found. Therefore, the incidence of malignant teratoma among all ovarian malignant tumors was approximately 6.2 per cent compared with the 1 to 4 per cent of other larger series (Diddle, 1949; Montgomery 1948). The apparent divergence in the incidence may be partly explained by the sampling of a small series.

Since benign cystic teratoma is a totipotent tumor, any type of malignant degeneration or a combination of malignancies can be expected. Among these, the most frequent type is squamous cell carcinoma. Other less common malignancies arising in benign cystic teratoma of the ovary are adenocarcinoma, sarcoma, endothelioma, perithelioma, choriocarcinoma and carcinoid (Peterson, 1957). In both of our cases the ovarian cyst was composed of ectodermal structures, and no mesodermal or entodermal elements were encountered, inspite of the multiple sections examined microscopically, the malignant degeneration was squamous cell carcinoma.

An interesting fact was the coexistance of two different malignant tumors; a squamous cell carcinoma arising in benign cystic teratoma of the right ovary and a serous cystdenocarcinoma in the opposite ovary, observed in one of our cases (Case 2). Although malignant degeneration of a benign cystic teratoma is itself a rare complication, a combination such as this case is more rare. Recently, Sharma et al. (1960) reported a case having three genital tumors in one woman a granulosa and theca cell tumor of the ovary, endometrial carcinoma of the uterus and squamous cell carcinoma of the cervix. Also a case

of choriepithelioma occurring in one ovary and malignant disembryoma of the other ovary was reported by Marion et al. (1957). In this connection, Siegler and Silverstein (1947) claimed support for the theories of ectopic origins in pathological entities such as these when they reported a case with both a dermoid and an endometrial cyst.

In our two cases, malignant degeneration was not suspected in the benign cystic teratomas until sectioning the tumor, even though the cyst was densely adherent to neighbouring structures at one area in one of our cases (Case 1). Therefore, in the treatment of these benign cystic teratomas, two points appear to warrant emphasis: 1) The possibility of malignancy should always be kept in mind in an apparently benign cystic teratoma, and one or more sections should be taken from the dermoid plug or mamilla, which is the common site of malignancy, in the operating room. In addition, any thickened, nodular or papillary area in the cyst wall calls for careful investigation. 2) Aspiration of these cystic tumors to ease the removal of the tumor without rupture may offer a better prognosis, since it was well pointed out in Peterson's paper (1957) that rupture of the cyst during operation gave a poor prognosis. However, aspiration of the characteristic content of a dermoid cyst has been found to be difficult in our hands. Accordingly, the technique of removing the content of the cyst should be modified.

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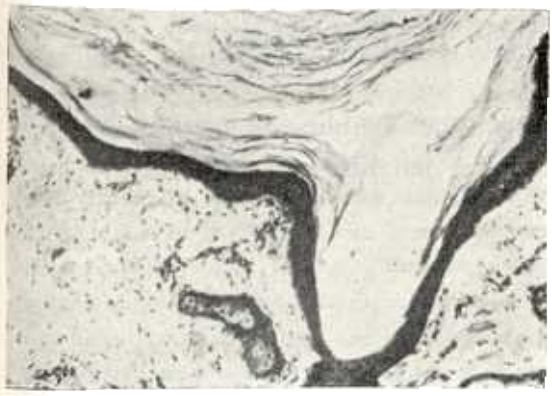

Fig. 1. (Case 1). Cyst wall showing epidermal lining, sebaceous glands and laminated layers of keratin. Hematoxylin and eosin stain. ×100

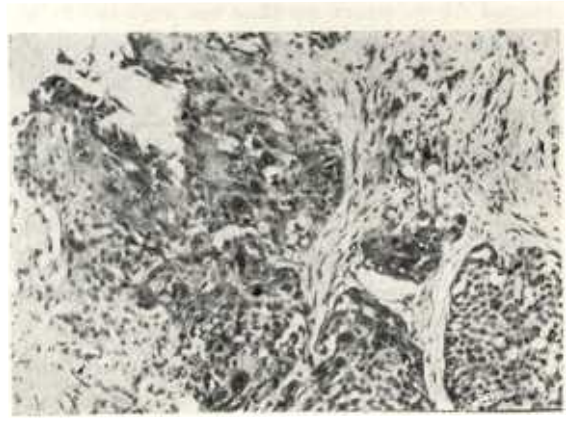

Fig. 2. (Case 1). An area of malignant degeneration composed of nests of neoplastic squamous cells. Hematoxylin and eosin stain. ×100.

Fig. 3. (Case 2). Cyst wall showing epidermal lining, sebaceous glands and laminated layers of keratin. Hematoxylin and eosin stain. ×100

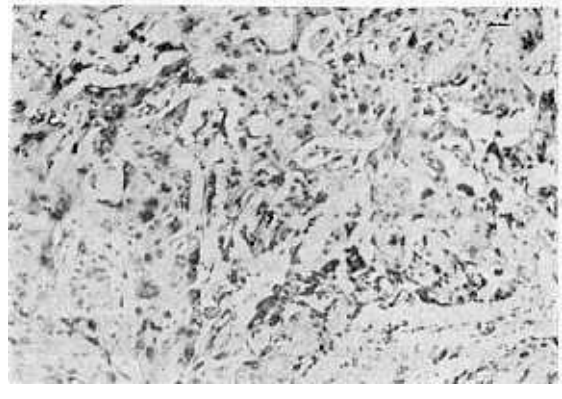

Fig. 4. (Case 2). An area of malignant degeneration showing infiltrating nests and cords of neoplastic squamous cells. Hematoxylin and eosin stain. ×100.

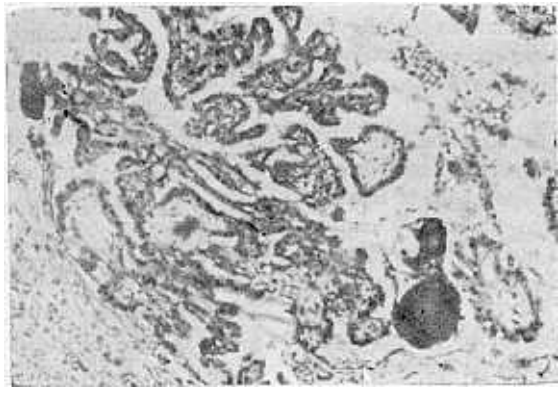

Fig. 5. (Case 2). Microphotograph of left ovary showing papillary serous cystadenocarcinoma. Note psammoma bodies in the right lower portion. Hematoxylin and eosin stain. ×100.