Squamous cell carcinoma arising in mature cystic teratoma of the ovary: A case report

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Introduction

Mature cystic teratoma (MCT) is one of the most common ovarian tumors [1]. In contrast, squamous cell carcinoma arising in MCT, is extremely rare. It comprises approximately 1% of all ovarian cancers [2]. A case of a well-differentiated squamous cell carcinoma arising in MTC, diagnosed in a middle-aged woman, is presented and issues regarding the diagnosis and management of this rare complication are discussed.

Case Report

In the second half of December 1999, a 66-year-old woman was admitted to our Hospital suffering from acute abdominal pain. Two months prior to entry to the hospital she suffered from atypical abdominal pain and she also noted urinary frequency and an increase in chronic constipation. The pelvic examination revealed a large palpable mass filling the pelvis, particularly on the left side. The rest of the physical examination was noncontributory. Ultrasonographic examination revealed a large, semicyst mass 14x12 cm approximately, replacing the left ovary.

A conservative left salpingo-oophorectomy was perfomed and the cyst was removed intact.

Pathology report (2224/1999). The specimen was a firm irregular mass measuring 14x12x0.3 cm. The external surface was white and smooth. The internal surface was multiloculated and a nodule of whitish-gray tissue, 3 cm in diameter, projected into the lumen. The wall of the specimen contained an area of thickening that measured 6x4 cm and was composed of soft yellowish-gray tissue. The left tube was normal. A routine histological examination followed and sections were stained with haematoxylin and eosin.

Microscopic lesions from several parts of the specimen revealed it to be a mature cystic teratoma, lined partially by skin, containing hair follicles and sebaceous glands. Deeper in the wall of the cyst, cartilage tissue and respiratory-type pseudostratified columnar epithelium were seen. Microscopic sections from the nodule revealed an *in situ squamous cell carcinoma (Figure 1)* that showed limited and focal invasion of the underlying stroma which presented a considerable inflammatory reaction *(Figure 2)*.

Diagnosis: Mature cystic teratoma with in situ squamous cell carcinoma with limited and focal invasion of the stroma.

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Discussion

The mature cystic teratomas make up almost 20% of all ovarian neoplasms [1]. They are unilateral in 88% of all cases [1] and provoke symptoms relating to the mass (pain, urinary and gastrointestinal complaints and menstrual irregularities) [3]. Occasionally they are accompanied by hemolytic anemia [4].

Microscopically Blackwell *et al.* [5] found ectodermal derivatives in 100% of the tumors, mesodermal structures in 93% and endodermal derivatives in 71%. A female nuclear sex chromatin pattern is present in all cases; chromosomal analysis has shown a 46,XX pattern [6].

The most common malignant change in cystic teratoma is squamous cell carcinoma, which accounts for approximately 80% of the total [2], followed by carcinoid tumor and adenocarcinoma [7,8]. Other unusual types of malignancy are melanoma [9], sarcoma of various types and thyroid carcinomas [2].

The malignant component of squamous cell carcinoma arising in MCT sometimes is observed in only part of the lesion, as in our case, causing difficulty in the preoperative diagnosis of malignant transformation of MCT.

Benign cystic teratomas have been seen in all age groups, the peak incidence being in the third and fourth decades. The mean age at the time of diagnosis is 37.5 years [10]. On the other hand malignant changes have been seen in greatest incidence in the fifth and sixth decades, as in our case and the mean age is 55.2 years [11].

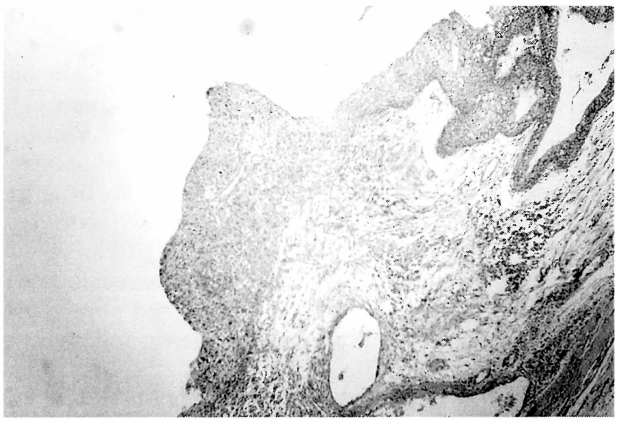

Figure 1. — Histological appearance of in situ squamous cell carcinoma arising in a mature cystic teratoma (haematoxylin and eosin x 250).

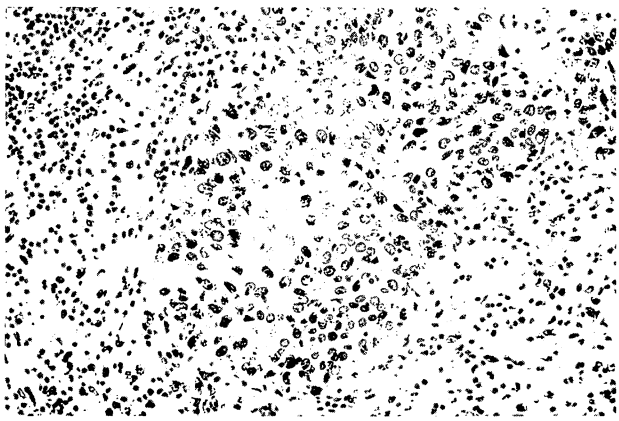

Figure 2. — Characteristic histological appearance of squamous cell carcinoma with focal invasion of stroma and inflammatory reaction (haematoxylin and eosin x 400).

Gross changes that suggest malignant transformation are adherence to surrounding structures, nodules or thickenings in the cyst wall, areas of hemorrhage or necrosis [8]. Tumor size is an important diagnostic factor. In general, malignant ovarian tumors are larger than benign tumors. Comerci *et al.* [12] analyzed 517 cases of MCT and reported that the mean tumor size was 64 mm, whereas Kikkawa *et al.* [10] analyzed 37 cases of squamous cell carcinoma arising in MCT and reported that the mean tumor size was 152.3 mm, as in our case.

Squamous cell carcinoma antigen (SCC) and carcinoembryonic antigen (CEA) until recently were considered rather poor diagnostic markers. Kikkawa *et al.* [10] suggest that CEA is the best screening marker for squamous cell carcinoma arising in MCT and SCC is the second best. Consequently both SCC and CEA serum levels must be examined if the patient is 45 years old or older and the tumor is more than 99 mm in the greatest dimension.

The generally accepted treatment for older women with limited spread of disease has been total hysterectomy and bilateral salpingo-oophorectomy [2]. For patients in the reproductive age group the therapy is less radical in some instances [8].

The prognosis of ovarian squamous cell carcinoma is much worse than that of other epithelial ovarian cancers and depends to a large extent upon the presence of tumor adhesions to surrounding tissues or rupture at the time of operation [8].

Patients with a squamous cell carcinoma arising in MCT, confined to the cyst and removed without spillage, have a 63% absolute 5-year survival [2].

Postoperatively our patient recovered quickly and is alive and symptom-free 12 months later without adjuvant therapy.

Conclusion

Malignant transformation of a mature cystic teratoma should be suspected if thickening of the wall, adherence to surrounding structures or necrosis is found at the time of the operation.

Age and tumor size are important factors and are useful in the differential diagnosis of MCT and squamous cell carcinoma arising in MCT.

References

[1] Rosai J.: "Ovary". *Ackerman's Surgical Pathology*, Raven Press, 1996, volume 2, 8th edition, 1500.

[2] Peterson W. F.: "Malignant degeneration of benign cystic teratomas of the ovary: a collective review of the literature". *Obstet. Gynecol. Surv.,* 1957, *12*, 793.

[3] Peterson W. F., Prevost E. C., Edmunds F. T., Hundley J. M, Morris F. K.: "Benign cystic teratomas of ovary; clinico-statistical study of 1,007 cases with review of the literature". *Am. J. Obst. Gynec.*, 1955, *70*, 368.

[4] Payne D., Muss H. B., Homesley H. D., Jobson V. W., Baird F. G.: "Autoimmune hemolytic anaemia and ovarian dermoid cysts. Case report and review of the literature". *Cancer*, 1981, *48*, 721.

[5] Blackwell W. J., Dockerty M. B., Masson J. C., Mussey R. D.: "Dermoid cysts of the ovary. Their clinical and pathologic significance". *Am. J. Obstet. Gynecol.*, 1946, *51*, 151.

[6] Rashad M. H., Fathalla M. F., Kerr M. G.: "Sex chromatin and chromosome analysis in ovarian teratomas". *Am. J. Obstet. Gynecol.*, 1966, *96*, 461.

[7] Hirakawa T., Tsuneyoshi M., Enjoji M.: "Squamous cell carcinoma arising in mature cystic teratoma of the ovary. Clinicopathologic and topographic analysis". *Am. J. Surg. Pathol.*, 1983, *13*, 397.

[8] Kelley R. R., Scully R. E.: "Cancer developing in dermoid cysts of the ovary". *Cancer*, 1961, *14*, 989.

[9] Ueda Y., Kimura A., Kawahara E., Kitagawa H., Nakanishi I.: "Malignant melanoma arising in a dermoid cyst of the ovary". *Cancer*, 1991, *67*, 3141.

[10] Kikkawa F., Nawa A., Tamakoshi K., Ishikawa H., Kuzuya K., Suganuma N. *et al.*: "Diagnosis of squamous cell carcinoma arising from mature cystic teratoma of the ovary". *Cancer*, 1998, June 1, *82*(11), 2249.

[11] Stamp G. W., McConnell E. M.: "Malignancy arising in cystic ovarian teratomas: a report of 24 cases". *Br. J. Obstet. Gynaecol.*, 1983, *90*, 671.

[12] Comerci J. Jr, Licciardi F., Bergh P. A., Gregori C., Breen J. L.: "Mature cystic teratoma: a clinicopathologic evaluation of 517 cases and review of the literature". *Obstet. Gynecol.*, 1994, *84*, 22.

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