Bilateral primary squamous cell carcinoma of the ovary: A case report of isolated metastasis to the lateral pelvic wall

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Summary

Primary squamous cell carcinoma of the ovary is rare. The majority of cases arise most commonly from the lining of a dermoid cyst, and less often in endometriosis or a Brenner tumor. A 40-year-old woman underwent exploratory laparotomy and was found to have a right ovarian tumor adherent to the lateral pelvic wall with no ascites. She underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, pelvic lymphadenectomy, infracolic omentectomy, appendectomy, and right nephrectomy for bilateral primary squamous cell carcinoma of the ovary. She was started on multiagent chemotherapy. On follow-up after two years the patient had died of cerebral metastases.

To our knowledge in this report we present the first case in the English literature of bilateral pure squamous cell carcinoma of the ovary.

Key words: Squamous cell carcinoma; Ovary.

Introduction

Squamous cell carcinomas of the ovary arise most commonly from the lining of a dermoid cyst - less often in endometriosis or a Brenner tumor [1]. Metastases to ovaries from cervix uteri and undiscovered tumor foci of other organs have been reported [1, 2]. The literature also contains 24 cases of pure squamous cell carcinoma of the ovary with few or no clues to their origin [3].

In this report we present a case of bilateral pure squamous cell carcinoma of the ovary submitted to refractory radiotherapy and multiple chemotherapeutic agents with a poor outcome. To our knowledge this case is the first bilateral pure squamous cell carcinoma of the ovary to be reported.

Case Report

A 40-year-old white, multigravida woman underwent exploratory laparotomy and excisional biopsy for a right localised retroperitoneal tumor one year before at another hospital. The pathologic report revealed primary unknown squamous cell carcinoma. The patient received 50 Gy radiotherapy to the whole abdomen and pelvis. Because the tumor persisted the patient received different chemotherapy such as 5-FU, cisplatin, taxotere, and gemcitibine. However, her disease demonstrated no regression and the patient was referred to our hospital. She complained of right leg pain. Physical examination revealed that her right leg had edema. Bimanual pelvic examination demonstrated a normal sized uterus and cervix and a 6 x 7 cm firm adnexal mass. Computed tomography (CT) of the pelvis-abdomen revealed a 5 x 6 cm right-sided tumor in the pelvis, and non-functional right kidney. The level of serum CA-125 was 543 U/ml (normal: 0-35 U/ml).

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The patient underwent exploratory laparotomy and was found to have a right ovarian tumor adherent to the lateral pelvic wall and no ascites. The uterus, tubes, left ovary, omentum, and pelvic and periaortic lymph nodes were without clinical evidence of gross metastases. After cytological study, the right ovary was completed freed from the lateral pelvic wall using gentle dissection. Thereafter, she underwent total abdominal hysterectomy with bilateral salpingo-oophorectomy, pelvic lymphadenectomy, infracolic omentectomy, appendectomy, and right nephrectomy. There was no macroscopic residual tumor left following surgery.

Pathology

The right and left ovary measured 5 cm and 4 cm in diameter, respectively. Gross examination showed that both ovaries were occupied by solid and firm tumors, and infiltration to the wall of the uterine tubes and mesosalpinx was identified. Microscopic examination of both ovarian tumors revealed nests of malignant squamous cells in desmoplastic stroma. Tumor cells usually had scanty cytoplasms, but in some areas abundant keratinized cytoplasm and keratin pearls were identified (Figures 1 and 2).

Tumor cells showed reactivity for pancytokeratin (Pan-CK cocktail, Neomarkers) and cytokeratin-7 (CK-7, Neomarkers), but there was no reactivity for cytokeratin-20 (CK-20, Neomarkers). Histopathological findings were for other tissues - proliferative endometrium, chronic cervicitis, chronic pyelonephritis, reactive lymphoid hyperplasia and normal appendix. The case was diagnosed as bilateral squamous cell carcinoma of the ovary. Peritoneal washing cytology revealed no malignant cells.

The patient was put on taxol and carboplatin for three cycles. The level of serum CA-125 increased to 800 U/ml. Therefore she was replaced on topotecan 1.5 mg/m^2 x 5 days. She received three cycles of topotecan. However the level of serum CA-125 raised to 1204 U/ml and the patient experienced sudden unconsciousness. CT of the cranium demonstrated intracranial hemorrhage. She died of intracranial hemorrhage due to intracerebral metastases.

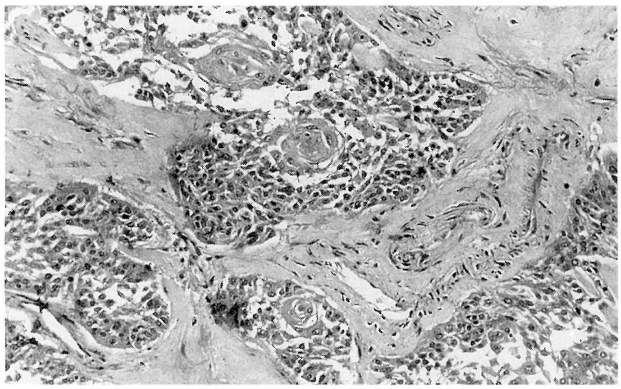

Figure 1. — Squamous cell carcinoma. Keratin pearls are seen in the nests of malignant squamous cells in desmoplastic stroma (Hematoxylin & eosin x 100).

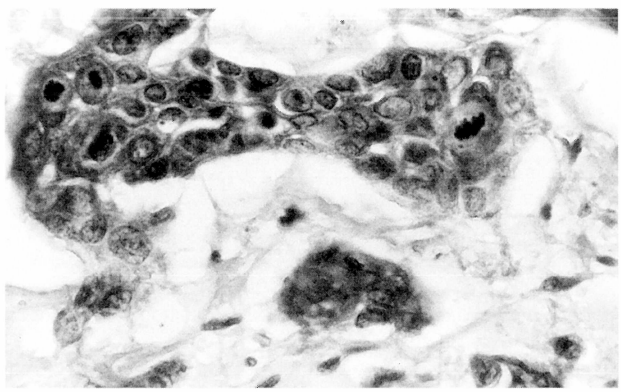

Figure 2. — Squamous cell carcinoma. Tumor cells have abundant cytoplasm (Hematoxylin & eosin x 400).

Discussion

Primary squamous cell carcinomas of the ovary must be distinguished from endometrioid adenocarcinomas with extensive squamous differentiation and from secondary cell tumors originating in the uterine cervix and possibly other sites [1]. Although primary squamous cell tumors of the ovary have been documented with dermoid cysts, endometriosis or Brenner tumor, extensive sampling from tumors - epidermoid cyst, Brenner tumor, Walthard nest, endometriotic foci, endometrioid adenocarcinoma, transitional cell carcinoma and teratomatous foci were not found. Vascular space invasion was not prominent.

It has been demonstrated that all primary ovarian carcinomas were keratin-7 positive and keratin-20 negative, with the exception of mucinous tumors [4]. In our case tumor cells showed strong positive reaction for CK-7; there was no reactivity for CK-20.

Involvement of the opposite ovary, as in our case, is a well known entity for primary ovarian carcinomas, and this situation may be a logical explanation for our case. Bilateral secondary ovarian involvement could be possible, but there was no evidence of distant organ (i.e. lung, breast) carcinomas showing CK-7 positivity. With these findings our case was diagnosed as bilateral primary pure squamous cell carcinoma of the ovary without other tumors.

Primary squamous cell carcinoma of the ovary is uncommon. Due to its rarity, there is no consensus regarding the effective treatment. Some authors have reported a poorer prognosis of patients with this type of tumor compared with other ovarian carcinomas [3, 5].

However, few studies have investigated the effect of paclitaxel and platin analogs [6, 7]. In these studies a remarkable response of primary squamous cell carcinoma of the ovary together with absence of significant side-effects related to therapy were reported. However their cases were associated with ovarian endometriosis or dermoid cyst.

Our case was diagnosed as a bilateral primary pure squamous cell carcinoma of the ovary with no other tumors. The patient received radiotherapy followed by cisplatin, 5-fluorouracil, taxotere and gemcitabine after the first operation. Because her disease showed progression tactual and carboplatin were started for three cycles after second debulking surgery, but there was no response. Recently, she was placed on topotecan, but she died of intracranial hemorrhage due to cerebral metastasis.

Her tumor was very resistant to chemotherapy and radiotherapy. She died of cerebral metastases two years after the first surgery in spite of intensive therapy such as radiotherapy and multi-agent chemotherapy.

References

[1] Scully R. E., Young R. H., Clement P. B.: "Transitional and squamous cell tumors". In: "Tumors of the ovary, maldeveloped gonads, fallopian tube, and broad ligament. Atlas of Tumor Pathology", Third series, fascicle 23, Rosai J., Sobin L. H. (Eds.), Washington D. C., 1988, 153.

[2] Young R. H., Clement P. B., Scully R. E.: "The ovary". In: "Diagnostic Surgical Pathology". Sternberg S. S. (Ed.) Third edition, Lippincott, Williams and Wilkins, Philadelphia, 1999, 2307.

[3] Pins M. R., Young R. H., Daly W. J., Scully R. E.: "Primary squamous cell carcinoma of the ovary. A report of 37 cases". *Am. J. Surg. Pathol.*, 1996, *20*, 823.

[4] Wauters C. C., Smedts F., Gerrits L. G., Bosman F. T., Ramaekers F. C.: "Keratins 7 and 20 as diagnostic markers of carcinomas to the ovary". *Hum. Pathol.*, 1995, *26*, 852.

[5] Kikkawa F., Ishikawa H., Tamakoshi K., Nawa A., Suganuma N., Tomoda Y.: "Squamous cell carcinoma arising from mature cystic teratoma of the ovary: A clinicopathologic analysis". *Obstet. Gynecol.*, 1997, *89*, 1017.

[6] Eltabbakh G. H., Hempling R. E., Recio F. O., O'Neill C. P.: "Remarkable response of primary squamous cell carcinoma of the ovary to paclitaxel and cisplatin". *Obstet. Gynecol.*, 1998, *91*, 844.

[7] Ohtani K., Sakamoto H., Masaoka N., Shimada K., Kanaeda T., Kurihara M. *et al.*: "A case of rapidly growing ovarian squamous cell carcinoma successfully controlled by weekly paclitaxel-carboplatin administration". *Gynecol. Oncol.*, 2000, *79*, 55.

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