A case of uterine cervical carcinosarcoma recurrence who obtained a clinically complete response by ifosfamide, doxorubicin and cisplatin

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Summary

Cervical carcinosarcoma (CS) is a rare gynecologic tumor. The histogenesis, clinical features, and optimal treatment remain unclear. We report a case of cervical CS recurrence to the right lung, which had complete response by treating with ifosfamide, doxorubicin and cisplatin (IAP). A 61-year-old woman underwent semi-radical hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy for CS of the uterine cervix. Eleven months later, the patient presented with left pulmonary metastasis. She refused debulking surgery and had chemotherapy with IAP. After four cycles of chemotherapy, the metastatic tumor completely disappeared. Unfortunately, a re-recurrent tumor was seen in the same lung area six months after IAP. Eventually, she died 39 months after surgery.

Key words: Uterine cervix; Carcinosarcoma; Recurrence; Chemotherapy; Clinically complete response.

Introduction

Carcinosarcoma (CS) is a compartatively rare gynecologic neoplasm which arises mainly in the ovary or uterine endometrium. CS represents around 3% of all uterine malignancies [1]. Because of the rare occurrence of this tumor, its histogenesis, clinical features, and optimal treatment remain unclear. Cervical CS is much rarer in gynecologic malignancies. We report a case of cervical CS recurrence to the right lung in which a clinically complete response was obtained by treatment with ifosfamide, doxorubicin and cisplatin (IAP).

Case Report

A 61-year-old postmenopausal woman, gravida 2, para 2, presented to our hospital with irregular vaginal bleeding of three months duration. Vaginal exploration showed a cervical polypoid mass occupying the vagina. The cervical mass was 6 × 5 cm on magnetic resonance imaging (MRI) T2 image, arising from the cervix, with no invasion of the vaginal wall. Neither remote metastasis nor hydronephrosis was noted on computed tomography (CT) and pyelography. The blood count and serum biochemical data and tumor markers such as CA125, CA19-9, SCC, CEA and LDH were within normal limits. Biopsy revealed cervical heterologous CS and FIGO Stage lb2. The patient underwent semi-radical hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy (Figures 1-a and 1-b). However, she did not have any adjuvant therapy since there was no stromal invasion and nodal metastasis according to the postoperative pathological examination.

Eleven months later, chest X-ray and CT scan showed a solitary metastasis in the area of the left lung and fine needle biopsy showed recurrence of the cervical CS (Figure 2, 3-a). The patient refused debulking surgery and had chemotherapy IAP. After four cycles of IAP, the metastatic tumor completely disappeared. Additional four cycles of IAP were given as consolidation therapy (Figure 3-b).

Unfortunately, a re-recurrent tumor was seen in the same left lung area six months after chemotherapy. Because of the dose limit of doxorubicin due to cardiac toxicity, the chemotherapy regimen was changed to paclitaxel and carboplatin (TC). Although partial remission was obtained by TC, she finally died 39 months after surgery.

Discussion

Uterine CS is a comparatively rare malignant neoplasm. The commonest site of occurrence is the body of the uterus. Cervical CS is extremely rare. The first case of cervical CS was described by Ferriera in 1951 [2]. There are only about 50 cases of cervix CS documented in the literature [3]. Most of these occur in postmenopausal women and form a polypiod mass, and the commonest clinical features are abnormal vaginal bleeding. Despite complete removal of tumor, the prognosis of CS is very poor and the metastasis rate is high [4]. Cervical CS is conventionally classified into homologous or heterologous as the CS originating in the corpus [5, 6]. The prognosis of heterologous CS may be better than that of the homologous [7, 8]. Complete surgical resection of the tumor most likely provides the best outcome of long-term survival. Radiotherapy to the pelvis reduces the risk of local recurrences, but there has been no survival advantage associated with postoperative radiotherapy [9, 10]. There are several negative opinions about adjuvant chemotherapy for uterine CS, whereas many more clinicians consider that chemotherapy for this tumor may be indispensable [3, 5]. Localized CS treated by surgery alone carries a high risk of local recurrence and metastatic disease. Of patients with clinical Stage I–II uterine CS

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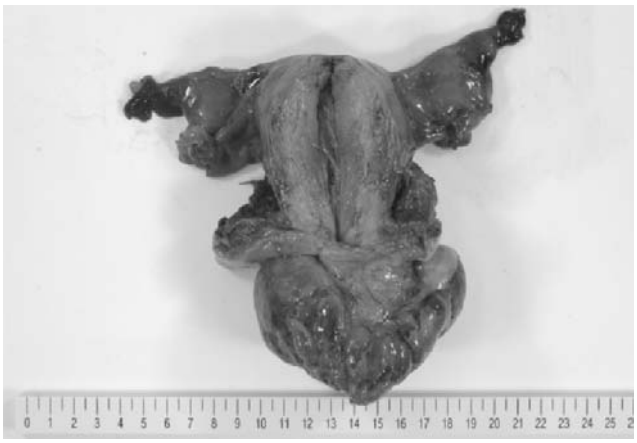
Fig. 1A



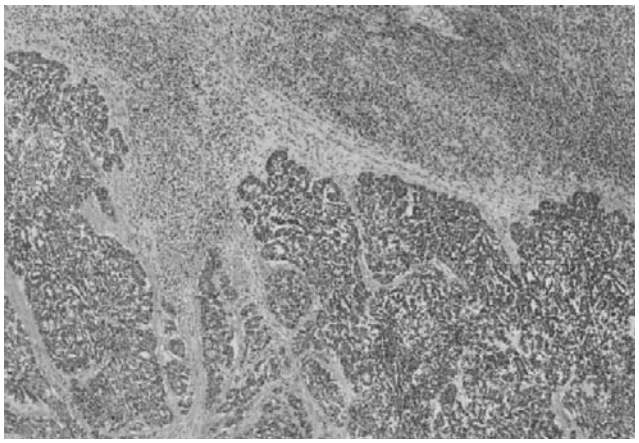


Fig. 1B

Figure 1. — A) Gross view of the surgical specimen. A polypoid tumor showing exophytic growth, measuring 9 × 9 × 4.5 cm. B) Histological section reveals malignant epithelium forming a glandular pattern. The background stroma also shows a sarcomatous pattern (original magnification × 40).

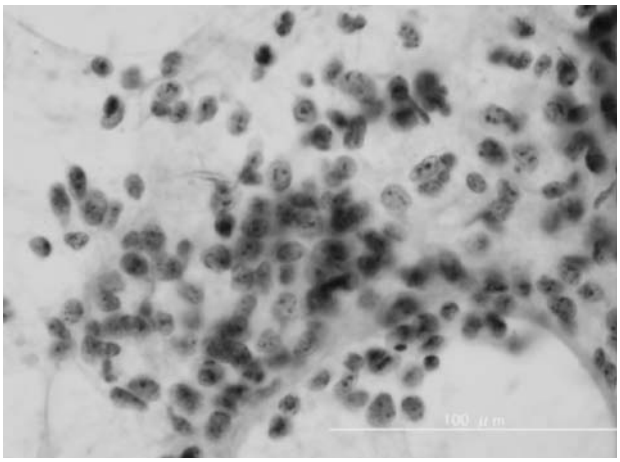


Figure 2. — Microscopic findings. Left lower lobe lung metastasis by transbronchial needle aspiration cytology (TBAC). Many malignant spindle cells can be seen (original magnification × 400).

53% developed recurrent disease within five years after total hysterectomy with surgical staging [11]. The single-agent doxorubicin or cisplatin or ifosfamide has activity against CS [12-15]. And the single-agent paclitaxel has had "moderate activity" [16]. The overall treatment response rate was 56% by chemotherapy with ifosfamide, doxorubicin and cisplatin [17]. The combination is superior to single-agent ifosfamide, doxorubicin or cisplatin in terms of response in patients with advanced, persistent and recurrent uterine CS [18-20]. That is why we chose ifosfamide, doxorubicin and cisplatin for this patient. However, toxicity of IAP combination treatment is not trivial. These drugs have myelotoxicity, doxorubicin cardiac toxicity, and cisplatin nephrotoxicity. Toxicity of this case was not severe; each was all within grade 2 in the National Cancer Institute Common Terminology Criteria for Adverse Events (CTCAE) ver. 3.0.

Future directions in the treatment of recurrent or metastatic CS of the uterus remain unclear. Also, the standard therapeutic method is not established at present. In this case, IAP therapy was very effective with mild toxicity and a clinically complete response. In other words it should be considered that IAP therapy can affect recurrence of CS of the heterologous tissue. However, no firm conclusions can be drawn because of the exceedingly rareness of such tumors.

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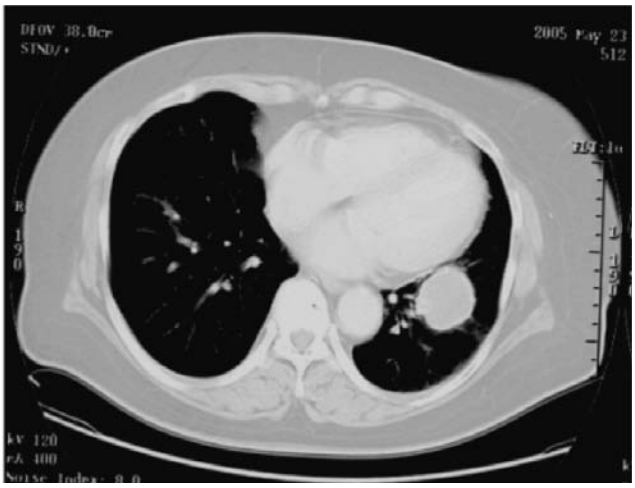
Fig. 3A



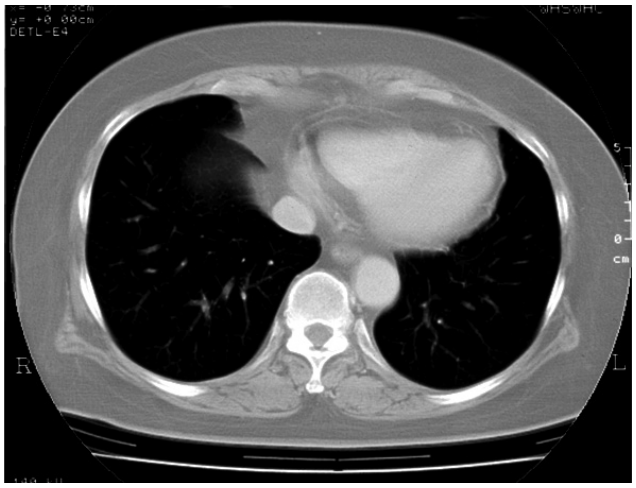


Fig. 3B

Figure 3. — A) Pretreatment CT scan showing 3 cm metastasis of the left lower lobe lung. B) CT scan showing complete disappearance of the left lower lobe lung metastasis.
There was no evidence of recurrence nor metastasis after the 6 courses of ifosfamide, doxorubicin and cisplatin.

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