Sarcomatoid squamous cell carcinoma of the uterine cervix: case report

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Summary
Sarcomatoid squamous cell carcinoma (SSCC) is a recognized entity, usually involving the upper aerodigestive tract and skin. Location in the lower female genital tract is rare. Only 11 cases have been previously reported, four of which arose in the uterine cervix.

The authors describe the case of a 39-years-old woman with SSCC of the uterine cervix in Stage I of the International Federation of Gynecology and Obstetrics (FIGO) classification. The patient died of disease 12 months after diagnosis.

Similar cases described in the literature are also reviewed.

Key words: Sarcomatoid Squamous Cell Carcinoma; Uterine Cervix.

Introduction
Cervical cancer is the most frequent gynecologic tumor in Portugal, excluding breast cancer, with a yearly incidence of 18 per 100,000 women. Squamous cell carcinoma is the most common histological type corresponding to 85% of all cervical carcinomas [1]. Other histological types are adenocarcinomas, sarcomas, melanomas, lymphomas, metastatic tumors and others [1].

Among squamous cell carcinomas different variants are recognized with prognostic implications. Sarcomatoid squamous cell carcinoma (SSCC) is one of the rarest variants of squamous cell carcinoma and it is characterized by its unique morphological features. Although it has a sarcomatoid appearance its epithelial origin can be demonstrated by immunohistochemical and by ultrastructural studies [2-5]. Usually two recognizable tumor components are evident: a well to moderately differentiated squamous cell carcinoma and a spindle cell component. Sometimes it is possible to identify areas of transition between both components [2, 3, 6].

This histological variant has been found in the oral cavity, pharynx, esophagus, larynx and skin but is seldom found in the female tract [2, 3, 5, 6].

Case Report
A 39-years-old caucasian female presented (in April 1998) with intermenstrual and postcoital bleeding of four months duration. A uterine cervix biopsy was performed and the patient was sent to the Gynecological Department of IPOFG, Lisbon, one month later.

The past medical and family history was unremarkable. In her clinical history, menarche was at 14 years old and she began sexual activity at 19 years of age. One sexual partner used coitus interruptus as the contraceptive method. The patient had one term pregnancy resulting in a healthy newborn.

On pelvic examination a friable and necrotic polypoid mass, approximately 6 cm in diameter, was found replacing the uterine cervix. The uterus was normal in size with a hard-elastic consistency and a smooth surface, and was freely movable. The adnexal areas, the vaginal fornices and parametria were free of tumor. The vagina and external genitalia were unremarkable. The histological diagnosis was sarcomatoid squamous cell carcinoma (Fig. 1). Blood chemistry was within normal limits except for the serum squamous cell carcinoma antigen (SCC= 12.7 ng/ml). Chest radiograph, abdominopelvic computed tomography (C.T. Scan) and abdominal and pelvic sigmoidoscopy were all negative for metastatic disease. The tumor was classified as Stage Ib (FIGO). Taking into consideration tumor size, she was scheduled for treatment with external pelvic radiation with 50.4 Gy (June, 1998). The patient was re-examined at 39.6 Gy and no clinical regression was seen suggesting absence of response to the treatment. Since the parametria were free of disease, the patient was submitted two months later to a radical hysterectomy with bilateral salpingo-oophorectomy.

The postoperative course was unremarkable. In the hysterectomy specimen a viable neoplasm was found with 7 mm at the greatest dimension. The patient was further treated with brachytherapy (50 Gy).

Nine months after diagnosis the patient occasionally referred pain in the right hip. The physical examination was negative and no clinical evidence of recurrent disease was found on pelvic examination. One and a half months later she had anorexia and weight loss, a productive cough with blood stained sputum and right chest pain. On physical examination two subcutaneous nodules, 3 and 5 cm in diameter, were found in the region of the right collar bone and shoulder. Again, on pelvic examination, there was no evidence of recurrent disease. The peripheral blood analysis showed microcytic and hypochromic anemia (Hb-5.6g/dl), leukocytosis (WBC-24.2x10⁹/l), decreased renal function (creatinin-1.8mg/dl, BUN-79 mg/dl), hypotremia (Na-124mEq/l), hypokalemia (K-2.4mEq/l) and hypercalcemia (Ca-15.4mEq/l). The chest radiograph demonstrated nodular opacities and the CT scan confirmed metastatic involvement of the lung as well as metastases in both kidneys, rib and right iliac bone (Figs. 2-5).

Fine needle aspiration of the subcutaneous nodules was performed for cytologic study and confirmed metastatic squamous cell carcinoma.

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Table 1. Sarcomatoid carcinoma of the uterine cervix.

<table>
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<tr>
<th>Case</th>
<th>Age</th>
<th>Stage</th>
<th>Therapy</th>
<th>Follow-up</th>
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| Steeper et al. [4] | 67  | Ib    | Partial excision, pelvic and para-aortic lymphadenectomy  
| Case 4    |     |       | Radiation and chemotherapy (int. after 1 week)                       | Died 2 months after diagnosis  
|           |     |       | Biopsy and radiation                                                   | (Intra-abdominal carcinomatosis)               |
| Steeper et al. [4] | 78  | III   | Radical hysterectomy and pelvic lymphadenectomy  
| Case 5    |     |       | Radiation and chemotherapy                                              | Died 7 weeks after diagnosis  
| Pang Leao C. C. [7] | 65  | Ib    | Chemotherapy and radiation                                              | Died 14 months after diagnosis  
| Case 1    |     |       | Died 12 months after diagnosis                                          | (multiple metastases)               |
| Pang Leao C. C. [7] | 61  | Ib    | Radiation, radical hysterectomy and bilateral salpingo-oophorectomy and chemotherapy | Died 12 months after diagnosis  
| Case 2    |     |       | Died 14 months after diagnosis                                          | (multiple metastases)               |
| This Report | 39  | Ib    | Radiation, radical hysterectomy and bilateral salpingo-oophorectomy and chemotherapy | Died 14 months after diagnosis  
|           |     |       | Died 12 months after diagnosis                                          | (multiple metastases)               |

Int. - Interrupted.  
- Metastasis in pelvic and/or para-aortic lymph nodes.

Figure 1. Viable spindle cell component in hysterectomy specimen (hematoxylin and eosin).
Figure 2. Thoracic CT scan shows several bilateral, nodular parenchymal lesions. The lesions are cavitated due to necrosis and the largest one has an air-fluid level.
Figure 3. Thoracic CT scan shows a well-defined extrapleural soft tissue mass in the right hemithorax which destroyed the adjacent rib.
Figure 4. Unenhanced abdominal CT scan demonstrates bilaterally enlarged kidneys with multiple foci of high and low density. The left kidney shows an area of infarction and absent function with an attenuation value near that of water (necrotic/hemorrhagic kidney metastases).
Figure 5. Pelvic CT scan: expansile lytic lesion of the right ilium with cortical destruction and an associated large soft tissue mass.
She was hospitalized and began chemotherapy with doxorubicin and vinorelbine (2 cycles). During the first cycle the patient had neutropenia and urinary infection, and during the second cycle she had neutropenia, thrombocytopenia and cardiomyopathy of the esophagus. Two weeks later the patient developed right hemiparesis. The brain CT scan revealed two metastatic lesions. Eleven months after diagnosis the patient's general status was rapidly deteriorating with anemia, dysphagia, cachexia, anasarca and worsening renal insufficiency. She died of the disease 12 months after initial presentation.

Discussion

Sarcomatoid squamous cell carcinoma (SSCC) is a recognized clinicopathologic entity in the upper aerodigestive, urinary tract and skin. The biological meaning of this entity is not fully understood.

Steeper et al. [3] and Enriile et al. [7] propose that this variant has a less aggressive clinical course in contrast with the more common squamous cell carcinomas of the respective sites.

Agha et al. [8] do not share this opinion. In their study of four cases localized in the esophagus, SSCC cases had a similar clinical course to “common” squamous cell carcinoma of the esophagus [8].

Few cases of SSCC have been described in the female genital tract [2-6]. In the lower female genital tract only 11 cases have been previously reported, four of which were located in the uterine cervix.

Analyzing all five SSCC cases located in the cervix and described in the literature including ours (Table 1), we found that all tumors occurred in postmenopausal women except the one we report, where the patient was 39 years old. The average age of all cases was 62 years. Clinically, the presentation was vaginal bleeding and/or a polypoid mass involving the cervix. Four of the cases were classified as Stage I, one case as Stage III, with regional lymph node involvement present at the time of diagnosis in three cases [3, 6].

Follow-up data was available in four cases and patients had a dismal outcome. Two patients died within the first three months after diagnosis and the other two died 12 and 14 months after initial presentation. Unusual metastatic sites were documented in both cases. Steeper et al. [3] described one case associated with peritoneal carcinomatosis and in our case distant metastases were documented, including, the kidney and subcutaneous tissue.

The different therapeutic management used does not allow us to draw definitive conclusions about adequate treatment for SSCC.

In conclusion, in spite of the small number of cases studied in this review, sarcomatoid squamous cell carcinoma located in the uterine cervix seems to have a more aggressive clinical behaviour in comparison to squamous cell carcinoma in which the five-year disease-free survival rate in Stage I tumors is around 85% of the patients [9].

Large series on this rare variant might determine the true biologic meaning of this entity.

References


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