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Recurrent cervical cancer presented with lymphangitic carcinomatosis

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Abstract

Lymphangitic carcinomatosis of the lungs secondary to cervical cancer is a rare condition. It still has an unknown pathophysiology and is related to high mortality rates. Diagnosis is often delayed due to the common symptoms of nonproductive cough and dyspnea. There are only 10 cases described in the literature. Most of the reported cases received only supportive care due to low performance status. Only three reported patients received palliative chemotherapy. We describe a case that has successfully received platinum-based chemotherapy.

Key words: carcinomatosis, cervical cancer, chemotherapy, dyspnea, lymphangitic.

Introduction

Lymphangitic carcinomatosis of the lungs secondary to cervical cancer is a rare condition. Only 10 cases have been described since its first report by Buchsbaum[1] in 1970. It still has an unknown pathophysiology and is related to high mortality rates. We describe a case that has successfully received platinum-based chemotherapy.

Case Report

A 57-year-old woman was referred to our institution in October 2007 with a recent history of genital bleeding. She was previously submitted to a loop electrosurgical conization at another institution in 1998, due to a high-grade squamous intraepithelial lesion with free surgical margins. From 1998 to 2007 she missed her follow-up.

Pelvic examination showed a 4.5-cm ulcerated lesion of the cervix and biopsy confirmed a squamous cell carcinoma. The right parametrium was compromised at its proximal portion and the patient was classified as stage IIB cervical cancer (FIGO).

The patient was treated with chemoradiation, based on weekly cisplatin (40 mg/m^2), 54 Gy of pelvic external radiation and four insertions of 700 cGy high dose brachytherapy.

After 14 months of follow-up, she presented with fever and mild dyspnea. Interpreted as pneumonia, she received levofloxacin. Her white blood count was between the normal range and her HIV serology was negative. She had no locoregional evidence of disease. As the dyspnea maintained, she was admitted in our institution and the antibiotics regimen was changed to ceftriaxone plus clarithromycin. Chest computed tomography (CT) revealed a diffuse interstitial pattern with small pulmonary nodules and small right pleural effusion (not eligible for thoracocentesis) (Fig. 1). After 10 days, the dyspnea persisted and she developed a chest pain. Coronariopathy, endocarditis and pulmonary embolism were discarded. Another chest CT was performed, revealing bilateral pleural effusion.

An abdomen CT showed mild right hydronephrosis and retroperitoneal lymph node recurrence. Thoracocentesis was performed on both sides. Analysis demonstrated a transudate, with negative cultures and

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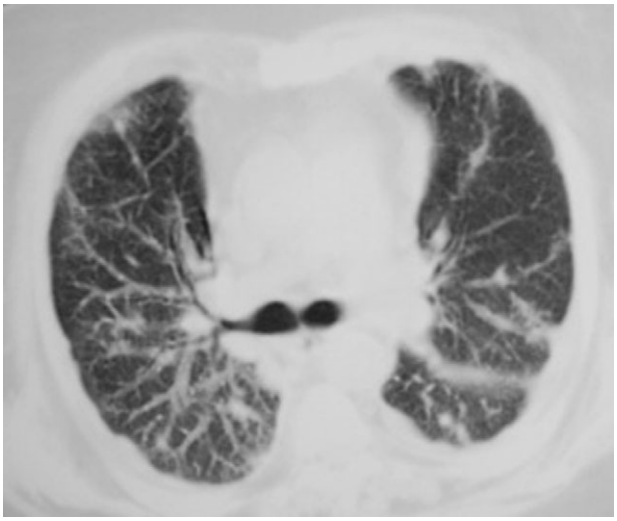

Figure 1 Chest computed tomography showing diffuse interstitial pattern and small pulmonary nodules.

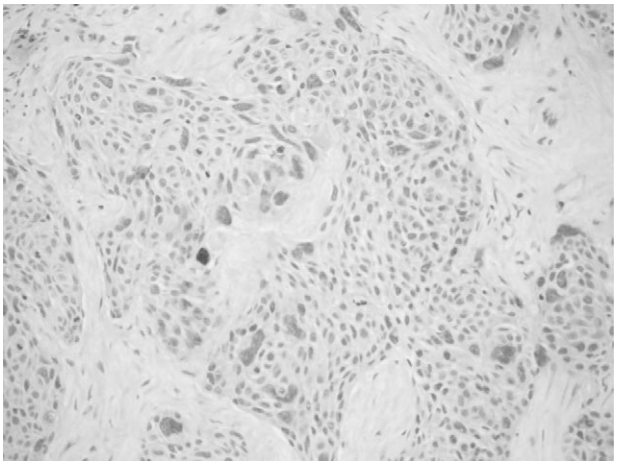

Figure 2 Microphotograph of metastatic carcinoma showing diffuse infiltration of pleural tissue.

cytology. Bronchoscopy with negative cytology and culture was also performed.

As the clinical status did not improve, a pulmonary and pleural biopsy through thoracotomy was performed. The biopsy confirmed metastasis to pleura and lymphangitic carcinomatosis (Fig. 2).

The patient has received five cycles of chemotherapy based on carboplatin and paclitaxel every 21 days until August 2009. The hospital stay length was 56 days. She had clinical improvement, with complete resolution of dyspnea, fever and chest pain after the third cycle. CT also showed partial pulmonary and retroperitoneal response. However, on October 2009, pleural and bone disease progression were diagnosed based on CT scans. She is still alive after 7 months of recurrence diagnosis.

Discussion

Pulmonary lymphangitic carcinomatosis is a rare event in gynecological malignancies. It is more common when secondary to other primary sites, such as stomach (most common), breast, pancreas, gallbladder, prostate, thyroid and larynx.[1] Regarding squamous cell carcinoma of the cervix, only 10 cases have been reported in the literature.[2] The cervical cancer is known for metastasizing to surrounding tissues and lymph nodes rather than distant sites, contributing to the rarity of this event.[3] Pulmonary metastasis secondary to cervical cancer ranges from 4% to 6%,[4,5] with an unknown rate of lymphangitic carcinomatosis. Shin *et al*.[4] showed a 3% incidence of reticulonodular pattern of metastasis in chest imaging, but without histological confirmation.

Diagnosis is often delayed due to the common symptoms of nonproductive cough and dyspnea. Our patient also presented fever, which led to an infectious disease hypothesis. This type of misleading diagnosis also occurred in other reports, as four (40%) patients were treated initially for pneumonia.[1,3,6] Chest imaging is highly characteristic, showing diffuse infiltrates that extend towards the periphery,[3] but is not pathognomonic. The preferred diagnosis method used in some reports was transbronchial biopsy.[2,3,6–8] In our report, we opted for thoracotomy because of previous normal bronchoscopy and pleural effusion.

The etiology of this condition is yet unclear. Ziedman *et al*.[9] suggest that compromised hilar lymph nodes could block the lymphatic draining and allow retrograde lymphangitic spread. Other authors proposed mechanisms such as vascular dispersion and accumulation of malignant cells in regional lymphatic vessels[10] and incompetent valves with aberrant lymphatic flow.[11] Another hypothesis is that radiation therapy may lead to pulmonary metastases by disrupting the physical barrier function and immunological surveillance mechanisms of lymph nodes.[3]

Most of the reported cases received only supportive care due to low performance status. Storck *et al*.[2] reported a median time from diagnosis of cancer to lymphangitic carcinomatosis of 360 days, with 17 days of median survival. Only three of all described cases were treated with chemotherapy.[1,6,8] Lipmann's patient

was the only with prolonged survival and also the only one who received cisplatin based chemotherapy (associated to bleomycin, oncovin and mitomycin).[8] The patient reported by Perez-Lasala *et al*. was treated with paclitaxel, and died after 60 days.[6] Buchsbaum described a 17-day survival with no described chemotherapy schema[1]. Our patient received three cycles with a carboplatin and paclitaxel regimen, and is still alive after 4 months. Corticosteroids seem to provide temporary relief and diminish both bronchiole inflammation and dyspnea, but have no long-term effect. In our subject, only a few doses of hydrocortisone were used during the treatment and were related to a good dyspnea improvement.

Acknowledgment

The authors declare that there are no conflicts of interest.

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