PATHOLOGICA 2016;108:148-150

Case report

Unusual pleural effusion from vulvar squamous cell carcinoma: report of a case and review of the literature

S. ERRA[1], G. PATRUCCO[1], G. SPERANZA[1], S. BARBERO[2], F. BRUCCULERI[2], G. TAVERNA[2]
[1] SOC Anatomia Patologica ASL AL, P.O. Casale Monferrato; [2] SOC Radiologia Diagnostica e interventistica ASL AL, P.O. Casale Monferrato (AL), Italy

Key words

Vulvar squamous cell carcinoma • Mesothelioma • Differential diagnosis • Immunohistochemistry

Summary

Vulvar tumors are not very common and account for about 4% of all cancers affecting the female genital organs. Frequently, malignant neoplasia of this site have squamous phenotype and the rare cases of metastasization are reported in the locoregional lymph nodes and in the surrounding organs.

We report a case of metastasization of a vulvar squamous cell carcinoma in an unusual place such as the parietal pleura, in a relapsing patient that was submitted to a surgical vulvectomy the previous year.

Introduction

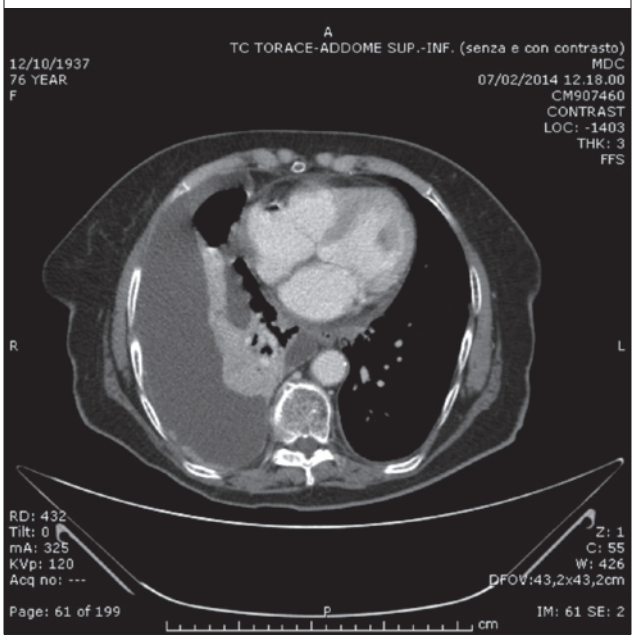
Carcinoma of the vulva accounts for 4% of total gynecological malignancies [1]. Tumors of the vulva can arise in women of all ages, even if the most cases are reported in postmenopausal patients and it represents a rarity in subject under 40 years, with an incidence of 1:100.000 in younger women, and 20:100.000 in the elderly [2]. Among the various histotipes of vulvar neoplasia, the most common is represented from squamous cell carcinoma [3].
Squamous cell carcinoma can arise from actinic keratoses or intraepithelial displasia on viral aetiology. Clinically it can be confused with other benign squamous lesions, such as warts or tumors of cutaneous appendages [4]. For this reason differential diagnosis is made on histological sections only. Squamous cell carcinoma is a locally aggressive tumor. It can have infiltrative activity towards subepithelial connective tissue and it can metastasize at the regional lymph nodes, interesting close organs too.
We report a case of metastatic squamous cell carcinoma in an unusual place represented by the right parietal pleura, in a relapsing patient submitted to a radical vulvectomy for a squamous carcinoma the previous year.

Case report

A 76-year-old woman presented in our hospital with respiratory symptoms, such as dyspnea and right chest pain. She was submitted to chest radiograph, evidencing the presence of monolateral pleural effusion, responsible to right atelectasis of the lower portion of the lung. In a second time, she was carried out a CT scan, confirming a right pleural thickening with small chips at the level of the posterio-inferior parietal site (Fig. 1). with mediastinal lymph nodes swelling. Primary pleural disease was suspected. One of the most frequent primary pleural malignancy observed in our hospital is represented by pleural mesothelioma, due to the endemic exposure to asbesto fibers in Casale Monferrato. The absence of thoracic retraction, the uninvolvement of the mediastinal pleura and the absence of chest wall infiltration didn't confirm the first clinical suspicion. In addition, anamnestic patient's history reported a surgical vulvectomy for a squamous cell carcinoma.

Correspondence
Stefania Erra, SOC Anatomia Patologica, ASL AL, P.O. Casale Monferrato, viale Giolitti 2, 15033 Casale Monferrato (AL) - Tel. +39 0142 434894 - Fax +39 0142 434368 - E-mail: stefania.erra@virgilio.it

Fig. 1. Right pleural effusion associated with pleural thickening mainly small chips at the level of the posterior parietal pleura.



Our patient underwent to pleuroscopy to obtain a bioptic sample of the affected area (Fig. 2). The removed fragments were formalin fixed and paraffin embedded. Routine histological slides revealed a pleural location of epithelioid high grade neoplasia with a tendency to show focal squamous differentiation. Immunohistochemical analysis supported this diagnosis, revealing a positivity for CK 5/6 (Fig. 3) and p63 (Fig. 4) and negativity for calretinin, CK 7 and CK 8/18.
On agreed morphological and immunohistochemical data, a diagnosis of pleural localization from squamous cell carcinoma was made.

Discussion

In the literature, few cases of distant metastasis from vulvar squamous cell carcinoma are reported. Five cases of bone metastasis are mentioned [5]. Although, they can't be considered true distant metastases, but the infiltration of the surrounding bone tissue from primary adjacent tumor. Previous reports mentioned 7 cases of distant metastases from a primary tumor of the vulva, even if they were not squamous histotypes, rather than endodermal sinus tumor, rare neoplasia of germinal origin. These patients underwent radical vulvectomy and subsequent chemotherapy, but in few months lung and pleural metastases were found [6].
Our case can be considered a rarer event respect of the cases mentioned above and reported in literature. In fact no case of vulvar squamous cell carcinoma with this clinical course has been described. The peculiarity of this reported case consists in presenting pleural localization without loco-regional involvement in a kind of

Fig. 2. Routine section of pleural biopsy coloured with Ematoxiline-eosine (10X).

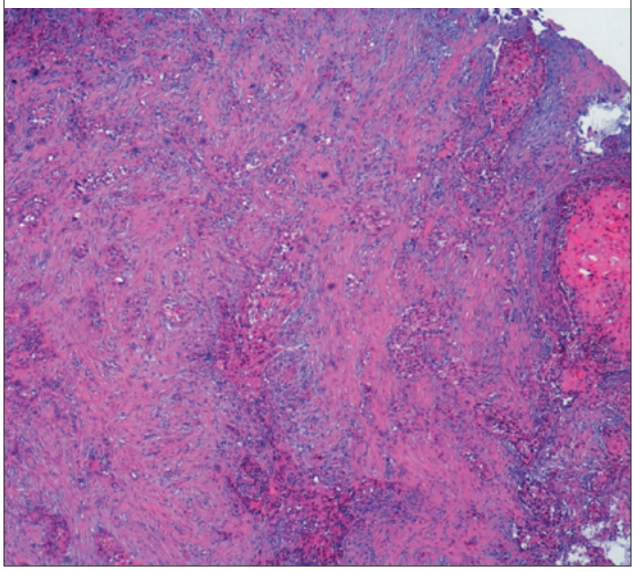


Fig. 3 . Immunohistochemical analysis revealed a positive immunophenotype for CK 5/6 (20X).

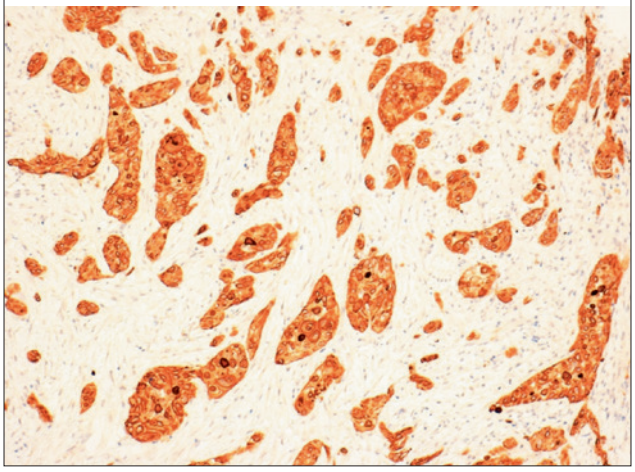
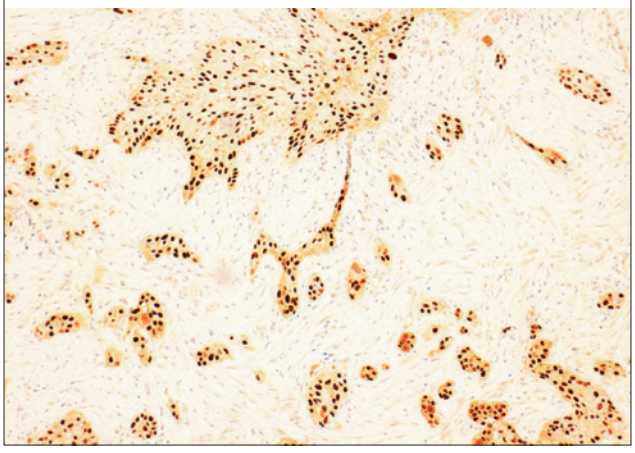


Fig. 4 . Immunohistochemical analysis revealed a positive immunophenotype for p63 (20X).



tumor known having locally aggressive course. Even if anamnestic clinical information can be considered important in the differential diagnosis, cases of more different malignancy in the same patient are not infrequent, for example in subject affected by hot-point genetic mutations or familial syndromes. For this reason, initial clinical symptoms suggested a pleural primitive tumor, supported by the fact that in population affering to our hospital incidence of mesothelioma is greater than national one. For diagnostic purpose, computer tomography thoracic scan and then histological picture of the lesion have been diriment. These exams allowed to discriminate a primary pleural tumor from a secondary localization of a neoplasia previously affected the patient. As in the most of cases, immunephenotypical characterization of tumor cells on formalin fixed-paraffin embedded bioptic samples is resulted indispensabile for the final diagnosis. This one is based on testing a panel of antibody to discriminate primitive mesothelioma from pleural secondary localization. In our experience, immunopositivity of neoplastic tissue for D2-40, calretinin, Wilms Tumor Gene1antibody and low molecular weight cytocheratin is suspected for mesothelial primitive neoplasia. This suspect is confirmed through the absence of expression of epithelial markers, such as Epithelial Membran Antigen, MOC-31, BerEp4 and CEA.

In our reported case, definitive diagnosis is based not only on the absence of expression of mesothelial antibodies, but on immunopositivity for antibodies detected in carcinoma wth squamous differentiation, such as p63. This evidence, together with the peculiar morphology of squamous cells, the anamnestic record of previous vulvar squamous carcinoma and, finally, the absence of strumental imaging suggesting other lesions in pulmonary district confirmed the pathology conclusion that vulvar squamous carcinoma can give distant localization without loco-regional involvement.

In conclusion, our case can be considered not only a rarity as regards biological behavior of tumors, but it should be considered an example of what a good clinical and basic medical history affects on both instrumental and morphological differential diagnosis of neoplastic lesions.

References

[1] Deka P, Barmon D, Shribastava S, et al. *Prognosis of vulvar cancer with lymph node status and size of primary lesion: a survival study*. J Midlife Health 2014;5:1-13.

[2] Tavassoli FA, Devilee P. *Tumor of the vulva*. In: *World Health Organization classification of tumors. Tumor of the breast and female genitale organs.* Lyon: IARC 2003, p. 316.

[3] Associazione Italiana per la Ricerca sul Cancro. Website, http//:www.airc.it (accessed November 4, 2014).

[4] Yamazato S, Nakai N, Katoh N. *A case of cutaneous squamous cell carcinoma arising in an actinic keratosis on the face in a centenarian woman successfully treated with surgery*. Indian J Dermatol 2014;59:304-6.

[5] Fischer F, Kuhl M, Feek U, et al. *Bone metastases in vulvar carcinoma: a rare metastatic pattern*. Int J Gynecol Cancer