UNILATERAL KAPOSI SARCOMA IN A PATIENT WITH PULMONARY TUBERCULOSIS AND HISTORY OF BREAST NEOPLASM

SARCOM KAPOSI UNILATERAL LA O PACIENȚĂ CU TUBERCULOZĂ PULMONARĂ ȘI ANTECEDENTE DE NEOPLASM MAMAR

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Abstract

Of all the four forms of the Kaposi Sarcoma, the iatrogenic form is the least encountered one. The occurrence of Kaposi Sarcoma in patients with a certified diagnosis of tuberculosis is thought to be a rare side effect of the delayed diagnosis of tuberculosis. What makes the below presented case special is the unilateral localization of Kaposi sarcoma and the breast neoplasm on the same side, the patient having been diagnosed also with active pulmonary tuberculosis.

Rezumat

Dintre cele patru forme ale sarcomului Kaposi, forma iatrogenă este cel mai puțin întâlnită. Apariția sarcomului Kaposi la pacienții cu tuberculoză este considerată o complicație rară a tuberculozei diagnosticate tardiv. Cazul prezentat este interesant prin localizarea unilaterală a SK și prin antecedentele de neoplasm mamar de aceeași parte, bolnava prezentând totodată tuberculoză pulmonară activă.
Introduction

Kaposi’s sarcoma (KS) is a vascular neoplasia with multiple expression at the muco-cutaneous level manifested through purple papules/nodules, with a marked tendency to growth and invasion and whose histological appearance associates vascular proliferations, spindle cells and lymphoplasmocytic infiltrate in varying proportions, depending on the evolutionary stage.\(^1\)

Kaposi’s sarcoma has been described by the dermatologist Moritz Kaposi, after having been observed on 5 male patients, under the following name: “multifocal pigmented hemangiosarcoma”.\(^2\)

Being a rare disease, it represents 0,02-0,08% of all the malignant tumors in Europe.\(^3\)

For many decades, KS has been considered as being a tumour of unknown origin, which affects mainly the elderly men from the Mediterranean countries, East Europeans and also the Jews. The interest for this disease started to increase in 1980s due to the high prevalence in those patients already diagnosed with HIV.\(^4\)

For a long period of time, it has been thought that KS has an infectious ethiology, but the attempts to demonstrate the main cause failed.

In 1994 Chang et al\(^5\) discovered a new type of herpes virus to an HIV infected patient. The newly found virus has been called Human Herpes Virus type 8 or HHV8 Kaposi sarcoma associated. The further undertaken studies demonstrated the presence of HHV8 in the majority of Kaposi lesions, no matter neither the clinical stage of the disease, nor the ethnity or the geographic origin. Thus, what strikes is the strong linkage between the KS and immunodeficiency, especially the cell mediated immunity, as well as the one observed to HIV diagnosed patients, patients with organ transplants or patients that have an iatrogenic suppression of the immunitary system. A few cases of KS in tuberculosis patients have been presented in the medical literature, KS having been considered a rare side effect of a delayed diagnosis of tuberculosis.

What makes this case special is the unilateral localization of KS, as well as the breast neoplasm on the same side, the patient having been diagnosed also with active pulmonary tuberculosis.

Clinical Case

F, 64 years, urban environment, requested dermatological consultation for eight purple colored, infiltrated tumors with a diameter of 0,5 cm and 2,5 cm, located in the right upper limb (Figure 1).

The onset was five months prior to admission, approximately a month after being diagnosed with active pulmonary tuberculosis and after the initiation of four anti-tuberculostatics therapy (isoniazide, rifampicine, pyrazinamide, ethambutol).

From personal history we retain that she had right breast neoplasm 19 years ago, treated by sectorectomy and right axillary lymphadenectomy and a duodenal ulcer in 2014. The general clinical examination has shown a patient with a good general condition, underponderal (Body Mass Index =18,3). In the right axillary region it could have been noticed the presence of a postoperative scar, having a diameter of 8 cm (Figure 2). Nonetheless, it has been noticed the absence of the superficial lymph nodes. The patient has been complaining that she started having, for approximately 6 months ago, sore cough.

The laboratory blood examination revealed normal values, excepting VSH (45/70 mm after an hour, respectively 2 hours).

An excision has been practiced, followed by a biopsy of a tumor, located to the right upper limb; the
Unilateral Kaposi Sarcoma in a Patient with Pulmonary Tuberculosis and History of Breast Neoplasm

Histopathological exam revealed a KS appearance (Figures 3,4). For the confirmation of the diagnosis, immunohistochemical examination has also been undertaken, which showed Vimentine, CD34, CD31, CD68 positive diffuse in tumor cells.

The immunological and imaging investigations that had been carried out, invalidated the breast cancer relapse diagnosis and confirmed the pulmonary tuberculosis evolution; the anti-tuberculosis therapy had been interrupted by the patient, on her own initiative, after 4 months of treatment. The investigations for the HHV8 infection had negative results.

Thus, based on the clinical and paraclinical examinations, the final diagnoses that were concluded are the following: Unilateral Kaposi Sarcoma; Active Pulmonary Tuberculosis; History of homolateral breast neoplasm.

Due to the lesions location (right upper limb), it has been chosen, as a treatment modality, the electrocautery.

**Discussions**

Kaposi Sarcoma can be classified in four forms: classical form, also known as the mediterranean, african or endemic form, AIDS-related Kaposi’s sarcoma and KS associated with iatrogenic immunodepression, the case that is the subject of this study also being included in this last category.

Kaposi’s sarcoma associated with immunosuppression, other than the one in AIDS, is a rare form of the disease. It is more commonly encountered in patients with organ transplants, having a global prevalence of 0.4% (8). KS lesions start to evolve, in average, after 16.5 months after the transplant; it’s possible also to have complete remissions if the immunosuppressive therapy is discontinued (7).

Cases of KS in patients with lymphoma under chemotheraphy and to those that were having a prolonged general corticotherapy have also been reported.

The extended immunosupression may represent a high risk factor, very important for the development of malignant skin tumors.

Patients with organ graft have a 65-250 folds higher risk to develop skin tumors compared to the general population. It is well known that the risk of developing a skin tumor is closely related to the fototype, the intensity of sun exposure, the age at the time of grafting and also, perhaps most importantly, to the degree of immunosuppression. (8)

In the general literature several cases of KS have been described, developed in patients undergoing immunosuppressive therapy, for various diseases such as pemphigus, bullous pemphigoid, dermatomyositis, Wegener’s granulomatosis, systemic lupus erythematosus, rheumatoid arthritis. (9,10,11) In a recently published article, two diagnosed cases with pemphigus vulgaris are described, which received immunosuppressive treatment with prednisone and azathioprime, KS lesions being developed in average after three months, respectively after three years after the initiation of therapy. (12)

In what concerns the origins of the KS tumoral cells, there are many uncertainties. Even though for a long period of time it has been known as a „sarcoma”, there is still a question whether the KS lesions represent a pure neoplasm or not. The studies published in the medical literature conclude to the assumption that the origin of the KS would be rather hyperplasic than neoplastic. (13,14)

The most recent and most disseminated theory related to the pathogenesis of iatrogenic KS suggests that, in the absence of a strong cell-level mediated immunity, the presence of the HHV8 virus might generate a spread of the infected endothelial cells, as well as a high increased risk and a certain genetic predisposition. (12)

Going back to the hereby case, it can be concluded that the neglected pulmonary tuberculosis had modified the imunologic status, which, by its own, increased a potential local imunosupression, generated by the right axillary limphadenectomy for the breast neoplasm, hence facilitating the evolution of the unilateral Kaposi sarcoma.

According to the above classification, the hereby case is being situated to stage I, which allows us to conclude towards a favorable evolution.

Regarding the KS treatment, in relation to the form and clinical stage of the disease, the major objective

### Table 1. The clinical stages of the Kaposi sarcoma (15)

<table>
<thead>
<tr>
<th>Stage</th>
<th>Features</th>
</tr>
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<tbody>
<tr>
<td>I</td>
<td>&lt; 10 skin lesions (limited to a single anatomic region)</td>
</tr>
<tr>
<td>II</td>
<td>&gt; 10 skin lesions (or extended to more than one anatomic region)</td>
</tr>
<tr>
<td>III</td>
<td>Only visceral ( node or gastrointestinal lesions)</td>
</tr>
<tr>
<td>IV</td>
<td>Skin lesions, visceral or pulmonary</td>
</tr>
</tbody>
</table>
| Subtype A | Without systemic signs or symptoms;  
| Subtype B | Fever > 37,8°C, unrelated to an identifiable infection, for more than 2 weeks; or weight loss > 10% of body weight. |
should be the symptom control. As treatment options we can mention: radiotherapy, surgery (for localized lesions), intralesional low-dose therapy with Vinca alkaloids or Bleomycin, cryotherapy, alone or in combination with other therapies, laser photocoagulation, hormonal manipulation therapy (using β-HCG), three therapy with antiproteases (AIDS-related KS). Each of the proposed therapeutic modalities has a variable success rate. Promising results were noticed after chemotherapy with cytotoxic agents (for extensive lesions and recurrence rate. Promising results were noticed after chemotherapy with cytotoxic agents (for extensive lesions and visceral involvement). The most recent publications on therapy with cytotoxic agents (for extensive lesions and visceral involvement). The most recent publications on therapy with cytotoxic agents (for extensive lesions and visceral involvement).

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**Conclusions**

Of all the KS forms, the iatrogenic form is the least frequent one, being encountered especially in patients with organ transplant, but also to those under immunosuppressive treatment for several pathologies. KS is a rarity among tuberculosis patients. Active pulmonary tuberculosis and right axillary lymphadenectomy for breast neoplasm have favored the development of Kaposi’s sarcoma located to the upper limb of the same side.

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**Bibliography**