Clinical and dermoscopic features of atypical abdominal brachytherapy-induced angiosarcoma

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Hemangiosarcomas are rare malignancies derived from blood vessel endothelium. In most cases, the origin of the tumor is difficult to be accurately determined due to poor cell differentiation. As far as skin is concerned, there are three clinical forms of the neoplasm:

1. Idiopathic hemangiosarcoma – most commonly located on the facial skin or scalp.
2. Hemangiosarcoma presented with limb edema in patients after mastectomy – referred to as Stewart-Treves syndrome.
3. Radiation induced angiosarcoma (arising in previously irradiated skin areas).

Angiosarcomas of deep soft and breast tissue in women were also reported. Recently an epithelioid angiosarcoma has been described as a rare aggressive variant [1–4].

Neoplasms, due to their clinical picture, are usually diagnosed late. Initially, the lesions appear as purple or red slow-growing patches. Nodules, which can undergo ulceration, appear over time.

Histopathological examination may reveal cell areas with polymorphic cells, numerous mitoses and irregular vascular channels. Some tumor cells have cytoplasmic vacuoles which correspond to the forming vessels. On the periphery, there are vascular channels with few atypical endothelial cells.

On dermoscopy, angiosarcoma is characterized by a combination of typical colors in vascular lesions – red, purple and blue. At the nodular part of the tumor, white lines can be detected. Radiation-induced angiosarcoma exhibits a more homogeneous pinkish-white pattern [5, 6].

Benign vascular lesions like rosacea, bruising, tumid lupus erythematosus or infections such as erysipelas and cellulitis are most often considered in differential diagnosis [7, 8].

The present case relates to a female patient with extensive angiosarcoma in the atypical location of the lower abdomen, which was treated locally over a long period of time, and dealt with as an erythematous spot of unknown etiology. The patient's history revealed brachytherapy due to endometrial cancer.

A 77-year-old patient exhibited her first skin lesions 5 months earlier; these suggested erythematous spots and were accompanied by severe itching. They were localized on the abdominal skin and were characterized by rapid growth. After 2 months, the lesions, bluish-violet at that time, progressed rapidly, and additional nodules and infiltrates appeared simultaneously. The patient history revealed brachytherapy due to endometrial cancer in 2002. The patient underwent a histopathological examination in an outpatient clinic in 2014 which revealed that the skin had numerous smaller blood vessels below the epidermis and within the dermis. The endothelium showed features of stimulation.

At the time of admission to the Department of Dermatology at Medical University of Silesia, lesions within the abdomen were well circumscribed from the surrounding skin. They reflected some bluish-purple infiltrates within which compacted dark-blue nodules were observed. The lesion dimensions were about 30 × 20 cm. The peripheral lymph nodes were not enlarged (Figure 1).

Laboratory results: erythrocyte sedimentation rate (ESR), complete blood count (CBC) with peripheral blood smear, iron levels, electrolytes, aspartate transaminase (AST), alanine transaminase (ALT), γ-glutamyltransferase (GGT), bilirubin, creatinine, urea, protein, protein, protein electrophoresis, creatine phosphokinase (CPK), aldolase, triglycerides, cholesterol, joint reactions (Latex-R, Waaler-Rose test, ASLO), general urinalysis and glucose levels were normal. Chest X-ray and abdominal ultrasound did not reveal any deviations from the norm. Dermoscopic image showed structureless red, purple-blue areas, white lines, yellowish clods on the periphery of some nodules

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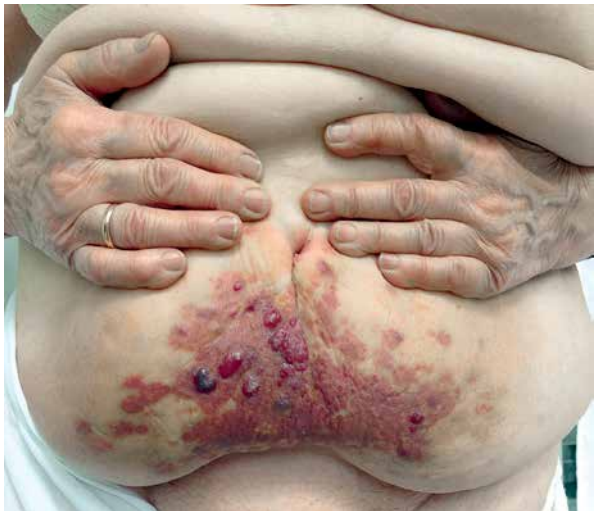

Figure 1. Lesions on the admission day – well-circumscribed bluish-purple infiltration with dark-blue nodules

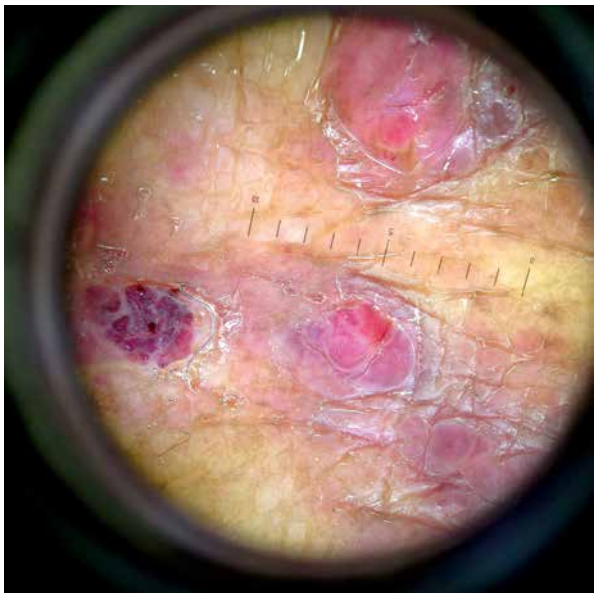

Figure 2. Dermoscopic image angiosarcoma – structureless red, purple-blue areas, white lines and structureless pinkish white areas and no solid pattern

and structureless pinkish white areas and no solid pattern (Figure 2).

Second histopathological examination confirmed the initial diagnosis: angiosarcoma. CD34+, Melan A–, LCA–, Ki-67+ in about 10% of cells.

External therapy, which slightly relieved the patient-reported pruritic ailments, was applied during the patient's stay at the clinic. The patient with histopathological diagnosis was referred to the Institute of Oncology for follow-up treatment. The therapy used palliative radiotherapy (the mean radiation dose was 70 Gy) – a few cycles of palliative chemotherapy with paclitaxel to render a slight improvement of the local condition. The patient was disqualified from surgical treatment. She currently remains under ongoing oncological observation for at least next 5 years.

Angiosarcoma is an aggressive malignant tumor of vascular endothelial cells or of lymphatic origin, whose clinical picture is variable and depends on the location. It occurs mainly in older white men. Its etiology is unknown. However, the effect of ionizing radiation is most frequently considered. The possible impact is also suspected in our patient's case.

Prognosis for cutaneous angiosarcoma, particularly in patients with tumors larger than 5 cm in diameter, is poor. The survival rate of 5 years is less than 30%. The treatment guidelines are not clear. However, surgical wide excision treatment with subsequent radiotherapy is basic. Yet, local relapses occur in 80% of cases [9, 10].

Although total surgical resection with subsequent radiotherapy is considered as the therapeutic standard for angiosarcoma, an appropriate choice of safe and effective adjuvant treatment is significant for elderly patients with comorbidities. Liposomal doxorubicin combined with radiotherapy seems to be an effective and safe option. However, all these therapeutic methods are generally palliative in nature and may only prolong the patient's life, rarely offering permanent recovery. This type of therapy was also applied for our patient [11, 12].

Angiosarcomas are usually fast-spreading tumors with a strong tendency to metastasize to the lymph nodes. Currently, our patient does not show any spread of the neoplastic process. The most rapid metastases occur in angiosarcomas located within the head and neck.

Histologically, angiosarcoma has variable features. Well-differentiated lesions may be misinterpreted as angiomas or lymphangiomas, while poorly differentiated lesions may mimic melanomas. Therefore, the final diagnosis should be confirmed by immunological examination. In the present case, the examination indicated angiosarcoma CD34+, Melan A–, LCA–, Ki-67+ in about 10% of the cells [13, 14].

Hung *et al.* [15] compared the clinical and biological characteristics of angiosarcomas of unclear etiology and post-radiation angiosarcomas (as in our patient). Average time between radiation therapy and diagnosis of angiosarcoma was 11.9 years (so it was close to our case, i.e. 10 years). The patients with angiosarcomas after radiation therapy for tumors other than breast cancer had a shorter survival time than in the case of breast cancer radiotherapy.

The present case relates to a female patient with extensive angiosarcoma of the lower abdomen, which was treated locally over a long period of time. The patient's history revealed brachytherapy due to endometrial cancer. The dermoscopic pattern although not pathognomonic for a specific diagnosis, was highly suggestive of a malignant tumour. Unfortunately, our patient's prog-

nosis is poor. She currently remains under ongoing oncological surveillance.

Conflict of interest

The authors declare no conflict of interest.

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