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Postoperative Scalp Angiosarcoma Was Accompanied by Ipsilateral Cephalic and Facial Herpes Zoster Appearance

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Abstract

Background: Cutaneous angiosarcoma is a rare cutaneous malignancy with poor prognosis. Because of its diverse clinical manifestations lack of specificity, clinical treatment is difficult.Here we present a case of postoperative scalp angiosarcoma with ipsilateral cephalic and facial herpes zoster appearance.

Case Report: An 88-year-old man was diagnosed with scalp angiosarcoma and underwent extensive excision and skin graft. Red induration occurred around the skin graft area of the scalp 1 month after surgery, along with ipilateral head and face blister-like changes and intermittent aggravated neuralgia. Finally, he died 3 months later due to refusal of further treatment. Immunohistochemistry was performed on sarcoma tissue and paracancer tissue obtained by pathological examination. The results indicated that the expression of NGF and TrkA were positive in sarcoma tissue and negative in paracancer tissue.It is speculated that there may be perineural invasion metastasis in angiosarcoma of scalp which is associated with sarcoma progression, increased local recurrence, intense pain, and poor prognosis.

Conclusion: Once cutaneous angiosarcoma patients indicate nerve invasion, immunohistochemical staining of pathological section NGF and TrkA should be performed when necessary to determine whether there is positive expression, so as to determine the possibility of perineural invasion metastasis. When such patients present with vesicular manifestations accompanied by progressive aggravated neuralgia, they should be highly vigilant about the possibility of sarcoma recurrence and perineural invasion metastasis, so as to actively take further treatment measures early to prolong the life of patients as much as possible.

Abbreviations

cAS: cutaneous angiosarcoma; PNI: perineural invasion; NGF:nerve growth factor; TrkA: tropomyosin receptor kinase A; CT: computed tomography; PET: positron emission tomography

Introduction

Cutaneous angiosarcoma((cAS)) is a rare cutaneous malignancy with poor prognosis. It is characterized by multifocal, diffuse, invasive, and high recurrence rate, and has the worst prognosis among all soft tissue sarcomas [1]. Various clinical manifestations of cAS have been reported in the literature, while herpes zoster appearance had rarely been mentioned, and they were all treated as herpes zoster in the early stage [2]. Based on this case, we speculated that angiosarcoma of scalp may have a metastatic mode of perineural invasion, which is associated with sarcoma progression, increased local recurrence, intense pain, and poor prognosis.

Case Report

An 88-year-old male with left top mass for 2 months and ulcerative bleeding for 1 week presented to our Plastic and aesthetic department on September 17, 2021.On physical examination, the mass on the top of the head was about 3*3.5cm, purple in color, medium in quality, unclear in boundary, poor in motion, red, swollen and ulcerated on the surface, a small amount of fluid seepage, and no obvious swelling of superficial lymph nodes in the neck (Figure 1a).

We suspected it was a skin malignancy. A biopsy was completed; pathology revealed vascular-derived tumors with heterotypic cells (considered angiosarcoma), immunohistochemistry was recommended (Figure 2).

Immunohistochemistry indicated that CD31(+), CD34(+), D2-40(+), ERG (+), CK (-), CK18(-), CD117(-), ki-67(50%+) (Figure 3a-d).

Cervical lymph node ultrasound did not indicate cervical lymph node metastasis (Figure 4a,b). Extensive excision of malignant scalp tumor and skin grafting was performed on the right upper arm were completed; pathology identified Hemangiosarcoma and all margins were negative with perineural invasion(Figure 5a-c).

Ten days later, the patient came to the hospital to have the stitches removed(Figure 1b).A staging PET/ CT scan revealed no systemic metastasis (Figure 6).

But after one week, the patient returned to our department for tingling pain on the left side of the head and face. On physical examination, he had multiple blisters appeared on the left head and face, the blisters were about the size of rice grains and the blisters were clear and clustered into sheets. Some of the blisters were broken

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Figure 1: Scalp clinical manifestations. a) Patient on admission; b) 10 days after surgery; c)17 days after surgery; d) 31 days after surgery.

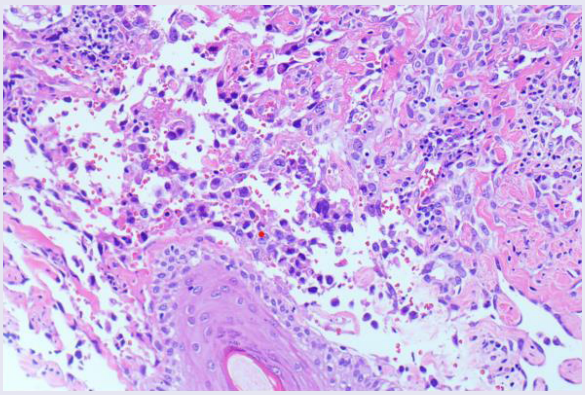

Figure 2: Scalp biopsy histology(200x). covered squamous epithelium, subcutaneous lumen with abnormal hyperplasia, anastomosis, enlarged endothelial nucleus, heterotypic, visible and schizotypic, incisal margin not clear.

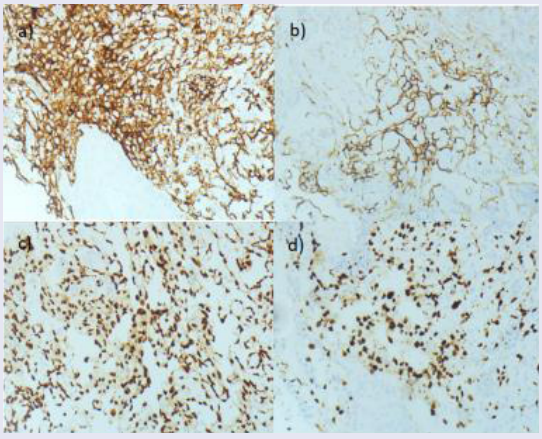

Figure 3: Scalp biopsy histology.a)immunohistochemical staining for CD31 demonstrating positive cytoplasmicr staining, indicative of Cutaneous angiosarcoma;b)immunohistochemical staining for CD34 demonstrating positive cytoplasmic staining ,indicative of Cutaneous angiosarcoma;c) immunohistochemical staining ERG demonstrating positive nuclear staining ;d) immunohistochemical staining for ki-67 demonstrating positive nuclear staining.

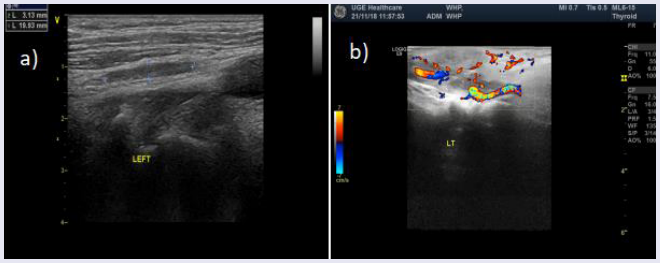

Figure 4: Cervical lymph node ultrasound showed that there were multiple low-echo nodes on both sides, with clear boundaries and capsula. The larger ones on the left and right sides were 20*3mm and 17*5mm respectively (a), And the blood flow signals in the nodules were not obvious; (b).

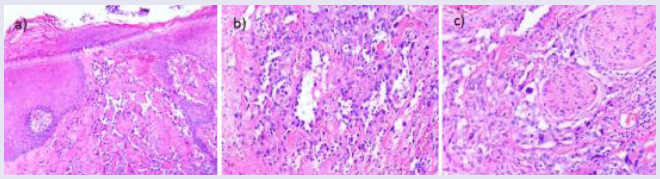

Figure 5: Postoperative scalp biopsy histology.a):Hemangiosarcoma of scalp forming a 3.5*2.5*0.6cm mass which covered with squamous epithelium, and the hypodermic dysplasia was observed(100x) ;b) Nucleolus could be seen, the mitotic image was easily seen, the lamellar arrangement was observed, and the abnormal hyperplasia lumen could be seen in some areas, which were consistent with each other (200x); c)All margins were negative with perineural invasion(200x).

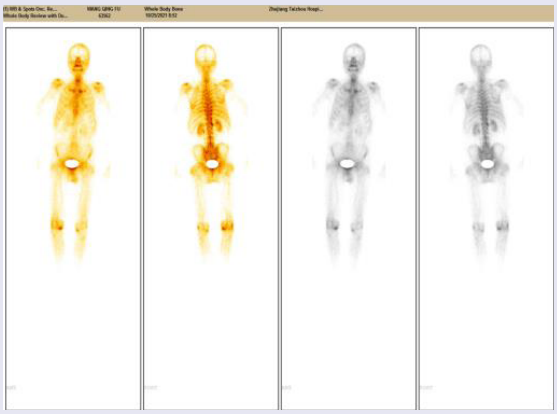

Figure 6: A staging PET/ CT scan revealed Bone metabolism of the left side of the parietal bone is mildly active(postoperative? Violation?)

and eroded, accompanied by a little exudation. The left eyelid was red and swollen, and a lump about the size of a coin was visible near the cheek of the left ear canal (Figure 1c). It compatible with herpes zoster were noted. Dexamethasone needles, penciclovir needles and mecobalamin tablets were initiated. His pain eased after 1 week of treatment.But after two weeks, the patient returned to our department for unbearable pain on the left side of her head and face. On physical examination ,multiple nodules with hard texture and poor mobility appeared at the margin of the skin graft area on the left top of the head (Figure 1d).We recommend needle biopsy of scalp induration to determine the nature of the scalp mass. The patient's family refused the treatment due to the patient's age to continue with

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morphine analgesic therapy.Follow-up 1 month later,the patient's family informed us that he passed away on December 23, 2021.

Discussion

Cutaneous angiosarcoma (cAS) has the worst prognosis among all soft tissue sarcomas due to its multifocal, diffuse, invasive and high recurrence rate [1]. cAS is very variable in clinical presentation and can present as hematomatoid lesions, but also as rosacea, eczema, hemangioma, purple spots or nodules, xanthoma, cellulitis, and angioedema of the face and eyelids.Nodules, papules, plaques and exophytic tumors appeared on the surface of advanced lesions [3]. If left untreated, these lesions grow rapidly and become multiple highly elevated nodular lesions with bleeding areas [1]. A variety of clinical manifestations of cAS have been reported in the literature, while shingle-like manifestations are rarely mentioned, and they are all treated as herpes zoster appearance at an early stage [2]. In the case presented here. The patient developed post-operative ipsilateral cephalic and facial herpes zoster appearance with intermittent worsening neuralgia, and peripheral red induration in the postoperative skin graft area. Subsequently, the patient developed red induration around the skin graft area after surgery. Combined with the advanced clinical manifestations reported in previous literature, it was considered that the recurrence of sarcoma still needed pathological support. The metastasis modes of cAS were mainly hematogenous (metastasis to lung, liver, bone), lymphatic metastasis, and local diffusion [4]. To date, PNI metastasis not been mentioned.By referring to relevant literature, we found that relevant studies on PNI in pancreatic cancer are relatively mature [5]. PNI is generally defined as the occurrence of tumor cells along the nerve and/or in the nerve sheath extraneuronal, extraneuronal, and intraneuronal, with cancer cells surrounding at least 33% of the nerve, and the survival time of patients with nerve invasion is significantly reduced compared with those without nerve invasion, and the risk of local recurrence and metastasis is significantly increased. Nerve growth factor (NGF) and its receptor TrkA are highly expressed in pancreatic cancer tissues, which can promote the proliferation and invasion of cancer cells and is associated with poor prognosis and cancer pain of pancreatic cancer [6]. Moreover, its high expression is more likely to find PNI in pancreatic cancer tissues [7]. The postoperative pathology of the patient indicated that nerve invasion (+). We consider recurrent and progressive manifestations of hemangiosarcoma from red induration around the skin graft area of the scalp, accompanied by ipsilateral cephalic and herpes zoster appearance due to PNI metastasis, and intermittent aggravation of neuralgia, resulting in a very short survival time.We used the monoclonal antibodies of NGF and TrkA ,and selected the sarcoma tissue and paracancer tissue from the patient for immunohistochemistry. The results indicated that the expression of NGF and TrkA in the sarcoma tissue was positive (Figure 7a) (Figure 8a), and the expression of TrkA in the paracancer tissue was negative (Figure 7b) (Figure 8b). It is speculated that there may be PNI metastasis in cAS, which is associated with sarcoma progression, increased local recurrence, intense pain, and poor prognosis.

Conclusion

cAS is a rare disease with poor prognosis,so the early recognition of this entity is essential to start treatment and reduce the risk of long-term sequela and even death.The cAS patient gave us the following tips:

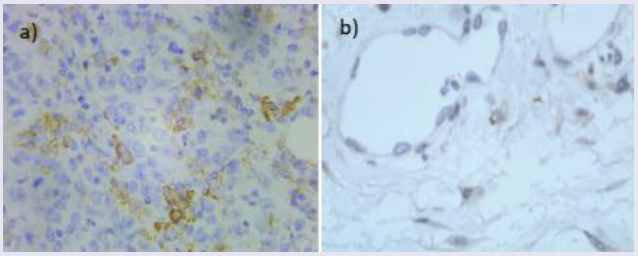

Figure 7: a) The sarcoma tissue: immunohistochemical staining for NGF demonstrating positive nuclear and cytoplasmicr staining; b) The paracancer tissue:immunohistochemical staining for NGF demonstrating negative nuclear and cytoplasmicr staining.

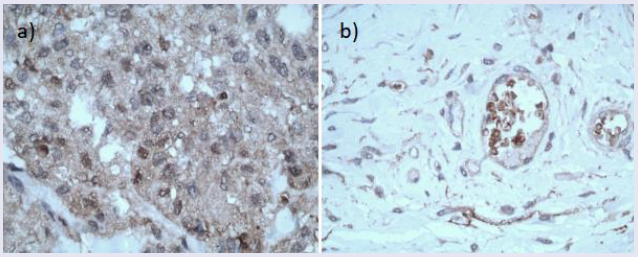

Figure 8: a) The sarcoma tissue: immunohistochemical staining for TrkA demonstrating positive cytoplasmicr staining;b)the paracancer tissue: immunohistochemical staining for TrkA demonstrating negative cytoplasmic staining.

(1) Once cAS patients indicate nerve invasion, immunohistochemical staining of pathological section NGF and TrkA should be performed when necessary to determine whether there is positive expression, so as to determine the possibility of PNI metastasis.

(2) When such patients present with vesicular manifestations accompanied by progressive aggravated neuralgia, they should be highly vigilant about the possibility of sarcoma recurrence and PNI metastasis, so as to actively take further treatment measures early to prolong the life of patients as much as possible.

In this case,as the patient did not undergo pathological biopsy around the skin graft area, it could not be determined whether the patient died due to local recurrence and metastasis, or the trauma caused by surgery, or other patient's own causes. However, during the whole process from diagnosis to final death of this patient, the local clinical manifestations of cAS still have certain clinical reference value.

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