The Journal of International Advanced Otology

J Int Adv Otol 2025; 21: 1–3 • DOI: 10.5152/iao.2025.241710

Case Report

An Extensive Bruise on the Hemiface: CARE Clinical Case

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Cite this article as: Rausch M, Quatre R. An extensive bruise on the hemiface: CARE clinical case. *J Int Adv Otol.* 2025, 21(1), 1710, doi: 10.5152/iao.2025.241710

Cutaneous angiosarcomas of the scalp are rare and aggressive tumors with non-specific appearances. They rarely affect the retroauricular region, and diagnoses are often difficult and delayed. We present the case of an 87-year-old patient referred for a large, spontaneously appearing, and rapidly progressing retroauricular bruise while on anticoagulants. Initial biopsies were negative, and the cervico-facial computed tomography (CT) scan with contrast injection was inconclusive. Despite stopping the anticoagulants, the lesion persisted and extended, leading to new biopsies that revealed a high-grade cutaneous angiosarcoma. Positron emission tomography-computed tomography (PET-CT) detected homolateral supraclavicular lymph node involvement and pleuropulmonary metastases. Given the patient's deteriorating general condition and the metastatic spread of the disease, exclusive comfort care was decided. With a highly variable clinical presentation, the diagnosis of cutaneous angiosarcoma is often delayed and made at a metastatic stage. Optimal treatment remains complete surgical excision followed by radiotherapy, but this is challenging in very extensive tumors.

KEYWORDS: Bruise, scalp, temporal bone, angiosarcoma

INTRODUCTION

Cutaneous angiosarcoma is a rare and aggressive vascular tumor that accounts for less than 1% of head and neck tumors.[1] More than 50% of cutaneous angiosarcomas are located in the head and neck region. This pathology most often affects elderly male subjects.[2]

The appearance of the lesion is often non-specific, with a bruised look that can be mistaken for a traumatic wound, hematoma, or benign lesion. With a very poor prognosis, the diagnosis is often made at a late or even metastatic stage, where surgical treatment is no longer feasible.

This pathology remains rare in ear, nose, and throat (ENT) consultations and is often mistaken for a spontaneous hematoma or one secondary to anticoagulant therapy. There are very few cases of angiosarcomas with a retroauricular onset reported in the literature.[3] Through this case, we aim to draw attention to this pathology, for which early diagnosis is crucial. This case study has been written following CARE guidelines (Case Report).

CASE PRESENTATION

An 87-year-old man was referred to our center for a right retroauricular bruise that had been evolving for 3 months. This patient was treated with oral anticoagulants (Rivaroxaban 20 mg) for a massive bilateral pulmonary embolism that occurred 7 years ago. The patient did not report any recent cranial or facial trauma.

The bruise initially occupied the right retroauricular region, with rapid extension to the ear, neck, and homolateral submandibular region. There was no visible ulcerated lesion, but some central hemorrhagic blisters were present (Figure 1). A first retroauricular

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Received: September 18, 2024 • **Revision Requested:** October 15, 2024 • **Last Revision Received:** October 21, 2024 • **Accepted:** October 30, 2024 • **Publication Date:** January 27, 2025
Available online at www.advancedotology.org

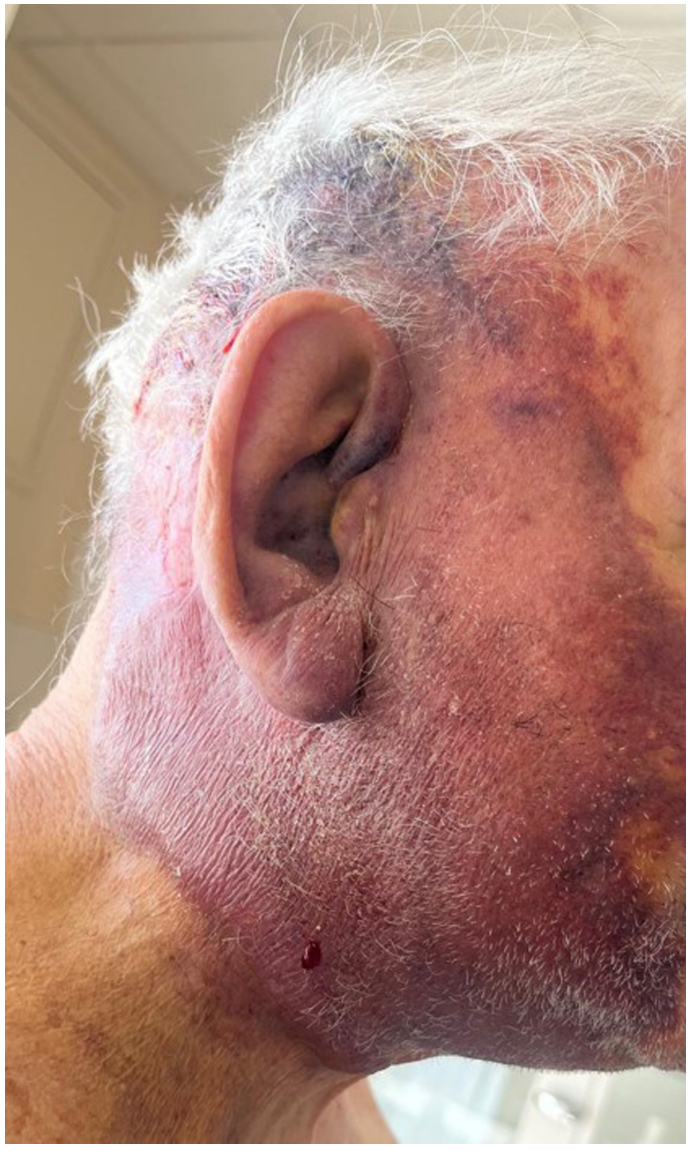

Figure 1. Clinical appearance of the retroauricular lesion with cervical and facial extension.

skin biopsy was performed under local anesthesia. The results suggested an ulceration with epidermal dystrophies without signs of malignancy.

In agreement with the hematologists, a biological assessment, including complete blood count (CBC), prothrombin time (PT), activated partial thromboplastin time (aPTT), fibrinogen, C-reactive protein (CRP), factors VIII, IX, XIII, von Willebrand factor, and venous occlusion time, was conducted to rule out a coagulation disorder.

Cervico-facial computed tomography (CT) with contrast injection revealed simple cutaneous and subcutaneous infiltration in the right retroauricular area, without arteriovenous malformation and without hematoma requiring surgical evacuation.

Given the extent of this bruise, anticoagulants were discontinued. Our initial hypothesis was a spontaneous temporal bone hematoma in the context of anticoagulation.

Subsequently, the bruise extended, with complete auricular and right temporofacial wall involvement.

Faced with this progression, and despite the discontinuation of anticoagulants, new retroauricular biopsies were performed, leading to the diagnosis of a high-grade retroauricular cutaneous angiosarcoma. Histological analysis revealed a tumor proliferation arranged in sheets with areas of highly atypical cells with hypertrophic, hyperchromatic, and nucleolated nuclei, along with mitotic figures. The presence of some vascular slits was noted. Immunohistochemistry showed intense and diffuse staining with CD31 and ERG markers. CD34 staining was faint and heterogeneous. The Ki67 proliferative index was high at 80%.

A positron emission tomography-computed tomography (PET-CT) scan was performed, revealing intense uptake in the entire lesion, as well as bilateral pleural metastatic effusion (Figure 2). Given the metastatic nature of the lesion and the patient's deteriorated general condition, chemotherapy seemed unreasonable. The patient was therefore offered exclusive comfort care.

The patient was informed and gave consent for this study.

DISCUSSION

Cutaneous angiosarcoma is a rare and aggressive vascular tumor predominantly located in the head and neck region. It develops from the walls of blood or lymphatic vessels. It can be secondary to irradiation or chronic lymphedema (Stewart–Treves syndrome).[4,5]

Presentation is variable and often non-specific, with violaceous bruised or nodular plaques on the scalp or face. Central ulceration or peripheral induration may occur. These lesions are often mistakenly considered benign skin lesions, leading to significant diagnostic and therapeutic delays. In our case, the diagnosis was made nearly a month after the first ENT consultation, and we initially suspected a spontaneous hematoma due to anticoagulants. Computed tomography scan with contrast injection, as well as magnetic resonance imaging (MRI), have low diagnostic yield for cutaneous angiosarcoma in the literature. The diagnosis is made through biopsy. It is crucial to repeat biopsies in the absence of clinical improvement, as they are often initially negative.[6] Histological diagnosis can be difficult in poorly differentiated forms, and it is not uncommon for initial biopsies to conclude with a benign vascular tumor.[5-7] Immunohistochemistry

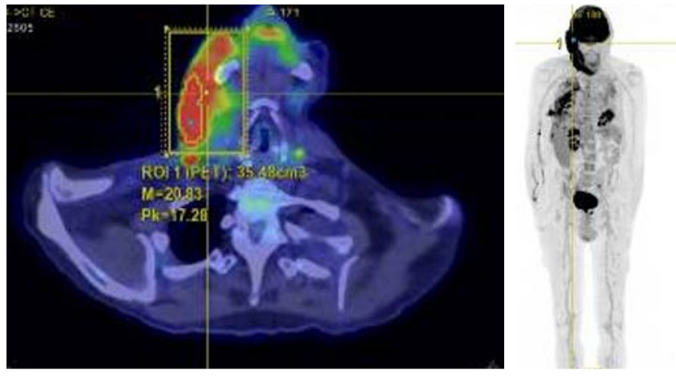

Figure 2. Positron emission tomography-computed tomography shows the hypermetabolism of the lesion and its local and distant extension.

often helps correct the diagnosis. The most commonly found markers are CD31 and CD34.[6] The rate of metastases is estimated at 36%, with the lungs, bones, and liver being the main affected sites.[7]

Despite the lack of official recommendations, optimal treatment involves surgical excision followed by adjuvant radiotherapy. A multimodal treatment combining surgery and radiotherapy seems to improve overall survival and reduce local recurrence rates compared to surgery alone.[2,8] There is no precise data on the resection margins required to limit local recurrences. Various series in the literature show a resection margin rate in healthy margins between 20% and 50% despite wide excision.[8-10] Paradoxically, negative resection margins do not always correlate with better survival.[9] Some studies instead recommend tumor reduction surgery with a simple and reliable reconstruction technique to avoid local complications and allow for the rapid initiation of radiotherapy.[9] In the case of very advanced and unresectable tumors, radio-chemotherapy or chemotherapy based on taxanes may be proposed as the first-line treatment.

According to different series in the literature, the 5-year survival rate varies from 33% to 54%.[2,7,10-13] The 5-year survival rate in patients who did not undergo surgery is estimated at 18% in a series of 107 patients.[14] The local recurrence rate is significant, up to 53%.[8] Tumor size is an important prognostic factor, with an improvement in overall survival noted in the literature for tumors measuring less than 5 cm.[15] Age also appears to be a major prognostic factor. In a cohort of 55 patients, Patel et al[2] showed that patients over 70 years old had a higher risk of recurrence. This is partially explained by the fact that elderly patients may have difficulty benefiting from multimodal treatment due to their deteriorated general condition.

CONCLUSION

Our clinical case demonstrates the diagnostic challenges posed by these cutaneous angiosarcomas of the face and neck, which are little known to ENT surgeons. This aggressive tumor manifested in this case as a simple retroauricular bruise in a patient on anticoagulants. This case also highlights the importance of repeating biopsies in the absence of clinical improvement in these patients.

Availability of Data and Materials: No data are associated with this article.

Ethics Committee Approval: This study was approved by the Ethics Committee of Grenoble AlpesUniversity Hospital (approval no.: 2205066 v 0, date: August 14, 2018).

Informed Consent: Verbal informed consent was obtained from the patient who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – R.Q.; Design – R.Q.; Supervision – R.Q.; Resources – M.R., R.Q.; Materials – M.R., R.Q.; Data Collection and/or Processing – M.R.; Analysis and/or Interpretation – M.R., R.Q.; Literature Search – M.R.; Writing – M.R.; Critical Review – R.Q.

Declaration of Interests: The authors have no conflicts of interest to declare.

Funding: The authors declare that this study received no financial support.

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