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Cheek Hemangioendothelioma : A Rare Localization of an Intermediate-Grade Vascular Tumor

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Abstract : Hemangioendothelioma is an intermediate-grade vascular tumor, and its occurrence in the cheek is exceedingly rare. This neoplasm often presents a diagnostic challenge due to its nonspecific clinical features and resemblance to other vascular or neoplastic lesions of the maxillofacial region.

We report the case of a 32-year-old female presenting with a slowly progressive, firm, and painless mass of the left cheek. Computed tomography and MRI revealed a well-circumscribed lesion with no evidence of bone invasion. An initial biopsy showed nonspecific fibroinflammatory tissue. Due to the persistence of the mass, surgical excision was performed via a paralateronasal approach. Histopathological examination of the resected specimen was consistent with an inflammatory myofibroblastic tumor. However, immunohistochemical analysis revealed a profile compatible with hemangioendothelioma.

This case highlights the importance of considering hemangioendothelioma in the differential diagnosis of facial soft tissue masses, and underscores the value of immunohistochemistry in establishing an accurate diagnosis.

Keywords : *Hemangioendothelioma, Cheek Mass, Vascular Tumor, Immunohistochemistry, Facial Soft Tissue Tumor.*

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I. INTRODUCTION

Hemangioendothelioma is a rare vascular tumor, classified between benign hemangiomas and malignant angiosarcomas. It encompasses a heterogeneous spectrum of lesions with variable biological behaviors, ranging from indolent progression to moderate metastatic potential. Among its various histological forms, localization in the soft tissues of the head and neck is exceptional, and even rarer in the cheek region [1,2].

This rarity, combined with nonspecific clinical presentation, poses a diagnostic challenge for both clinicians and pathologists. Indeed, hemangioendotheliomas can mimic other mesenchymal tumors such as myofibroblastomas, schwannomas, or fibro-inflammatory lesions [3,4].

Imaging may suggest a well-defined tumor, but it does not specify the lesion's nature. Thus, diagnosis relies primarily on histopathological analysis and immunohistochemistry, with frequent expression of endothelial markers such as CD31 and CD34 [5].

We report here an exceptional case of cheek hemangioendothelioma diagnosed in a young woman, initially mistaken for a fibro-inflammatory tumor. This case highlights the importance of broad diagnostic consideration and the use of immunohistochemistry in unusual facial masses.

II. CASE PRESENTATION

A 32-year-old female presented with a slowly progressive, firm, and painless mass in the left cheek. She reported no prior trauma or associated systemic symptoms. Clinical examination revealed a well-demarcated, non-tender swelling without overlying skin changes.

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Computed tomography (CT) and magnetic resonance imaging (MRI) demonstrated a well-circumscribed lesion located in the subcutaneous tissues of the left cheek, measuring approximately 58 × 30 × 70 mm, with no evidence of bone involvement or infiltration of adjacent structures.

An initial biopsy showed nonspecific fibro-inflammatory tissue. Due to persistence and progression of the mass, complete surgical excision was performed via a paralateronasal approach.

Histopathological examination of the resected specimen initially suggested an inflammatory myofibroblastic tumor. However, immunohistochemical analysis revealed strong positivity for endothelial markers CD31 and CD34, confirming the diagnosis of hemangioendothelioma.

Three months after excision, follow-up cervicofacial CT revealed a soft tissue process in the right maxillary region, suggestive of residual or recurrent tumor.

No surgical re-intervention was indicated. The patient underwent adjuvant radiotherapy with a total dose of 66 Gy delivered in 33 fractions.

Subsequent follow-up MRI revealed the emergence of a tumor centered on the right maxillary sinus, measuring 25 × 26 × 27 mm, associated with locoregional infiltration. Two suspicious lymph nodes, each measuring 10 mm in short axis, were noted in level II on both the right and left sides of the neck.

Weekly paclitaxel-based chemotherapy was initiated, resulting in a favorable clinical and radiological response. The patient remains under regular follow-up.

III. DISCUSSION

Hemangioendothelioma is a vascular tumor of intermediate malignancy, lying between benign hemangiomas and aggressive angiosarcomas. This entity encompasses several histological subtypes, each with variable biological behavior and relative rarity, particularly in the soft tissues of the head and neck, and even more so in the cheek region [6,7].

Jugular (cheek) localization of this tumor is exceptionally rare, making diagnosis particularly challenging. Clinical presentation is often nonspecific, typically as a painless, slowly enlarging mass, as illustrated in our case. Imaging can assess the localization and extent of the lesion, but remains insufficient for establishing a definitive diagnosis [6].

The differential diagnosis is broad and includes fibro-inflammatory tumors, inflammatory myofibroblastic tumors, schwannomas, and other benign or malignant vascular lesions [8,9]. The initial biopsy may be non-contributory, as in our patient, in whom it revealed a nonspecific fibro-inflammatory tissue.

Immunohistochemistry is an essential tool for confirming the diagnosis, through detection of endothelial markers such as CD31 and CD34. While these markers are sensitive, they are not entirely specific and must be interpreted in light of the histological and clinical context [10].

The management of hemangioendothelioma primarily relies on complete surgical excision, which remains the treatment of choice to minimize the risk of local recurrence. In cases of residual tumor or recurrence, adjuvant radiotherapy may be considered, as was done in our case [11].

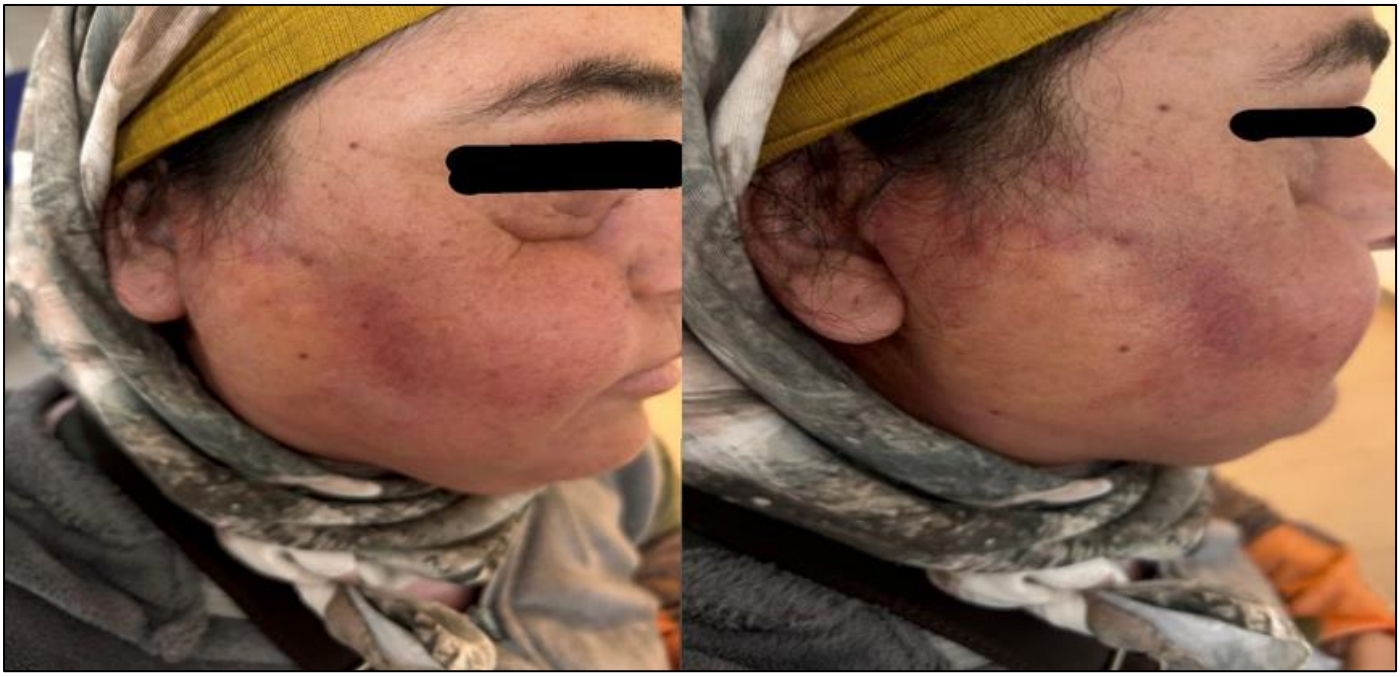

Fig1 Swelling of the Cheek with Local Erythema

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Chemotherapy, although not standardized for this entity, can be employed in situations of recurrence or locoregional extension. Protocols based on paclitaxel have shown some efficacy in this context [12].

Our case illustrates the diagnostic and therapeutic challenges posed by this rare tumor. It emphasizes the importance of a multidisciplinary approach combining imaging, histology, immunohistochemistry, and tailored treatments to optimize patient outcomes.

IV. CONCLUSION

Hemangioendothelioma of the cheek is an exceptionally rare vascular tumor that presents significant diagnostic and therapeutic challenges. Its nonspecific clinical presentation, combined with variable histological features, often leads to diagnostic delays. In our case, initial biopsy suggested a benign fibro-inflammatory lesion, underscoring the limitations of limited sampling. Immunohistochemistry, particularly the expression of endothelial markers such as CD31 and CD34, proved essential in confirming the diagnosis [10].

Given the intermediate malignant potential of hemangioendothelioma, complete surgical excision remains the mainstay of treatment [6,7]. However, in cases of residual disease or recurrence, adjuvant therapies such as radiotherapy [11] or paclitaxel-based chemotherapy [12] may provide clinical benefit. A multidisciplinary approach integrating imaging, histopathology, immunohistochemistry, and individualized treatment planning is critical to optimize outcomes in such rare cases.

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