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CHIRURGIE CERVICO-FACIALE

Disfiguring Intraparotid Angiolymphoid Hyperplasia with Eosinophilia: A Case Report

Braham R, Amri A, Zitoun O, Methnani A, Dhaha M, Jebali S, Dhembri S, Kedous S
ENT department, Salah Azaiez Institute, Tunis

Abstract

AIM : To report a rare case of a hemifacial vascular tumour, angiolymphoid hyperplasia with eosinophilia (ALHE), to study its clinical presentation, its imaging behavior, and its particular pathological aspects.
Methods : We studied the case of a 38-year-old female presenting with a hemifacial mass diagnosed as ALHE.
Case report : A 38-year-old healthy female patient was referred to our center for a soft-tissue tumor of the left cheek that had been increasing for over 3 years. Physical examination noted a House Brackmann grade 3 left peripheral facial palsy, a homolateral jugal polylobed mass extending to the paralateronasal region and filling the nasolabial fold, taking up the entire left hemiface with overlying plum-colored skin, with a 4cm submucosal mass on the inner surface of the left cheek. Magnetic resonance imaging revealed a left jugal subcutaneous fat-dependent tissue mass, infiltrating the superficial cervical fascia, coming into contact with the skin, isosignal on T1 and T2 sequences, with intense, homogeneous contrast, suggesting a fibrosarcoma. Exploration under general anesthesia via an incision along the left nasolabial fold revealed a diffuse white tumor adherent to the superficial planes and the canal of Stenon. An excision of the accessible portion of the lesion was performed in front of Bichat's ball. Histology was indicative of an angiolymphoid hyperplasia with eosinophilia. Despite the benign nature of the disfiguring lesion, the surgery was deemed invasive. Clinical monitoring was instituted to watch for extension of the vascular tumor and rule out other locations.
Conclusion : Angiolymphoid hyperplasia with eosinophilia is a benign acquired vascular neoplasm of unknown pathogenesis. Post-traumatic hypothesis with arteriovenous shunting is a suggested physiopathology. Diagnosis of certainty is based on pathology and should rule out Kimura’s disease.

Case report

38-year-old healthy female patient referred for a left cheek soft-tissue tumor growing for over 3 years.

Physical examination: House Brackmann grade 3 left facial palsy + left jugal mass extending to the paralateronasal region, filling the nasolabial fold, covering the entire left hemiface with plum-colored skin (figure 1)

MRI: left jugal subcutaneous tissue mass infiltrating the superficial cervical fascia, in contact with the skin, isosignal on T1 (figure 2) and T2 sequences (figure 3) and intense homogeneous contrast suggesting fibrosarcoma.

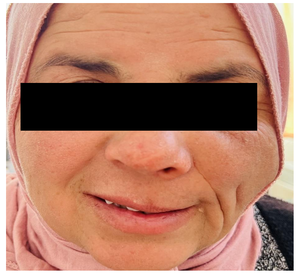

Figure 1 : left jugal mass with homolateral facial palsy

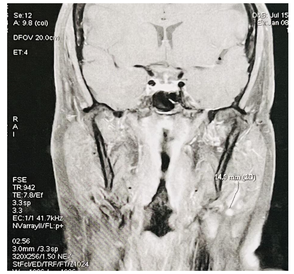

Figure 2 : T1 weighted MRI revealing isosignal left subcutaneous mass

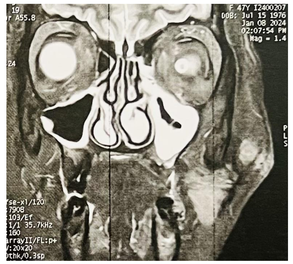

Figure 3 : T2 weighted MRI revealing isosignal left subcutaneous mass

Exploration under general anesthesia via an incision along the left nasolabial fold: diffuse white tumor adherent to superficial planes and the canal of Stenon.

Excision of the accessible portion of the lesion was performed in front of Bichat's ball.

Pathology: Angiolymphoid hyperplasia with eosinophilia; despite the benign nature, the surgery was invasive, and clinical monitoring was instituted to check for tumor extension and other locations.

Aim

Our aim is to report a rare case of a hemifacial vascular tumour, angiolymphoid hyperplasia with eosinophilia (ALHE), to study its clinical presentation, its imaging behavior, and its particular pathological aspects.

Conclusion

Angiolymphoid hyperplasia with eosinophilia is a benign acquired vascular neoplasm of unknown pathogenesis [1]. Post-traumatic hypothesis with arteriovenous shunting is a suggested physiopathology [2]. Diagnosis of certainty is based on pathology and should rule out Kimura’s disease [3].

Methods

We studied the case of a 38-year-old female presenting with a hemifacial mass diagnosed as ALHE.

References

[1] Alfarhan A, Maktabi A, Alofi MM, Alotaibi HM. Angiolymphoid hyperplasia with eosinophilia: a case series and literature review. Int Ophthalmol. 2023 Jul;43(7):2457-2467. doi: 10.1007/s10792-023-02644-y. Epub 2023 Mar 4. PMID: 36869976.[2] Guo R, Gavino AC. Angiolymphoid hyperplasia with eosinophilia. Arch Pathol Lab Med. 2015 May;139(5):683-6. doi: 10.5858/arpa.2013-0334-RS. PMID: 25927152.[3] Buder K, Ruppert S, Trautmann A, Bröcker EB, Goebeler M, Kerstan A. Angiolymphoid hyperplasia with eosinophilia and Kimura's disease - a clinical and histopathological comparison. J Dtsch Dermatol Ges. 2014 Mar;12(3):224-8. doi: 10.1111/ddg.12257_suppl. PMID: 24580875.