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Case Report

Arteriovenous malformation in cheek and tongue aggravated by cellulitis: A case report and literature review

Amine Kaouani [a,*], Ouassime Kerdoud [a], Rachid Aloua [a], Faical Slimani [a,b]

[a] Department of Stomatology and Maxillofacial Surgery, 20 August 1953 Hospital, Ibn Rochd, B.P 2698, Casablanca, Morocco
[b] Faculty of Medicine and Pharmacy of Casablanca, Hassan II University of Casablanca, B.P 5696, Casablanca, Morocco

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ABSTRACT

Arterio-venous malformation (AVM) is a congenital disorder of blood vessels, causing abnormal passage of blood between the artery and vein, which most often results in abnormal development of the vascular system in embryonic life.

Arteriovenous malformations of the head and neck pose a challenging therapeutic and reconstructive problem.

We present a rare case of a 22-year-old white woman, who complained of facial asymmetry on the right side, evolving after an episode of jugular cellulite of dental origin treated 6 months ago.

Clinical examination revealed a palpable nodule measuring approximately 25 mm, soft, painless, of submucosal location and a swollen, bluish right hemi tongue. Ultrasonography was requested, which indicated the hypothesis of a vascular lesion.

The microscopic diagnosis was of arterio-venous malformation with thrombus formation. The patient has been followed for 3 months and is without signs of relapse. We treated the cellulite and closely followed the patient.

1. Introduction

Arteriovenous malformations (AVMs) of the head and neck are rare and complex vascular lesions that can be present since birth with unpredictable growth. It is a structural vascular abnormality in which the arterial vasculature connects with the venous vasculature causing potentially disfiguring and life-threatening complications [1,6]. Infection, trauma, an ischemic event secondary to thrombosis or ectasia, hormonal changes such as pregnancy or puberty can induce AVM overgrowth [7]. The diagnosis is based on the history of the disease, the clinical examination, the imaging and lastly, if possible, the anatomo-pathological examination after surgery. The management of AVMs is very difficult, it must be multidisciplinary. Diffuse lesions have high rates of recurrent disease requiring vigilant follow-up and repeat interventions [2,4]. Our work is a single case report and has been reported in line with the SCARE criteria [3,20].

We report a rare and interesting case of a diffuse MVA of the right cheek and tongue.

2. Case report

A 22-year-old women, she has been followed since the age of 8 years for AVMs having benefited from a medical treatment based on beta-blockers. The symptomatology is of insidious appearance for several days. The patient has not benefited from any medical treatment. The patient noted some discomfort with severe localized pain associated with trismus. She was referred to our department's consultation for specialized care. No other personal or family history was raised during the patient interrogation.

She was admitted with a swelling of the right cheek progressively increasing in size with purulent endobuccal hemorrhage. Clinical examination revealed significant facial asymmetry, bluish swelling, reddish in places and fluctuating, pulsatile, the skin over her right face was remarkable with a prominent vasculature (Fig. 1).

The endobuccal examination reveals a limitation of the mouth opening to 3 cm. a swollen, bluish right hemilangue, from the base to the tip. The left hemi tongue is not swollen, but is bluish in places. The examination also reveals a very bluish inner face of the right cheek, with hemorrhagic pus on the lower vestibular side opposite the 48th tooth (Fig. 2).

* Corresponding author.
E-mail address: emaildeamine@gmail.com (A. Kaouani).

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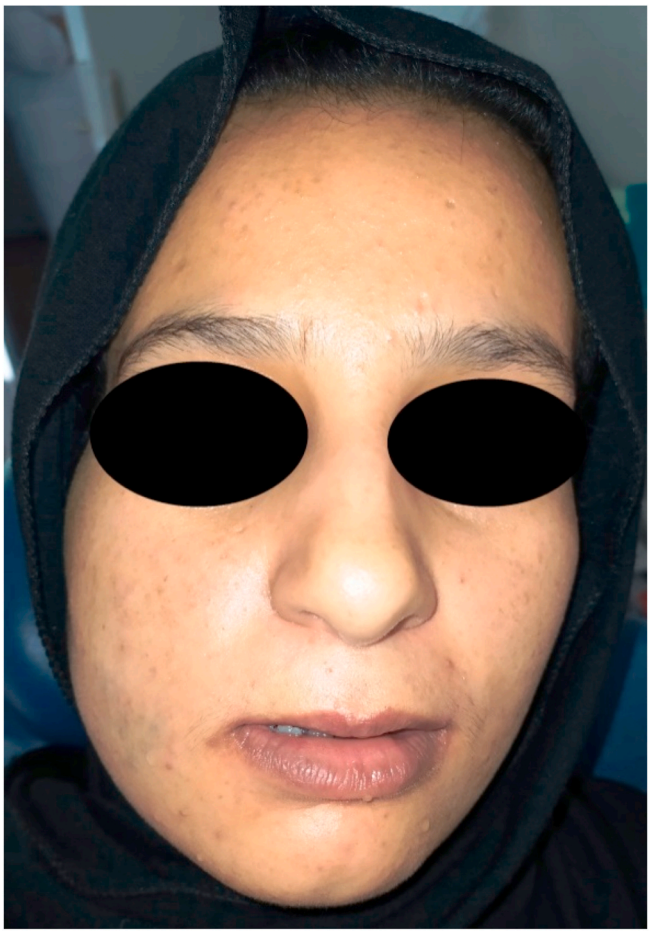

Fig. 1. Photo showing facial asymmetry, with right cheek swelling.

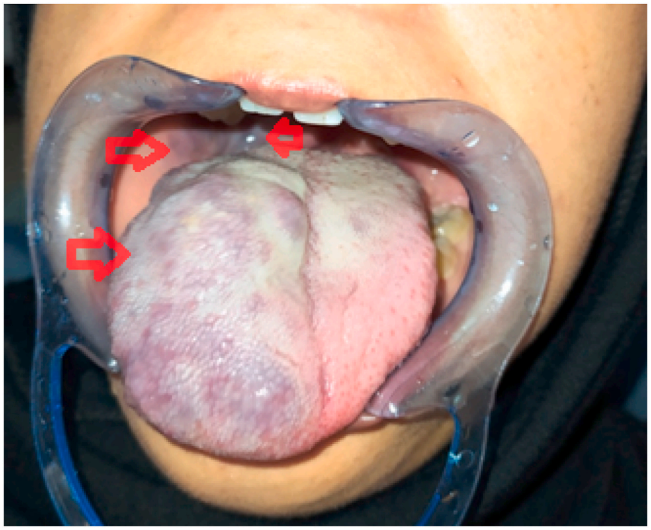

Fig. 2. Photo showing AVMs of the right cheek, right tongue and the pue.

The dental X-ray objectified opaque X-ray images at the area of the right hemiface. (Fig. 3).

The computed tomography (CT) was performed and revealeda cellulite collected on MAVs with multiple radiopaque lesions adjacent to the parotid gland consistent with phleboliths (Fig. 4).

The arteriography displaying tumor produced by a diffuse arteriovenous malformation fed by branches of the right external carotid artery. (Fig. 5).

A diagnostic of cheek cellulitis with AVMs was made based on the clinical examination and radiologic studies.

According the Suen-Richter classification system for arteriovenous malformations, the patient has Stage II: T2 D1-2 S1-2 [1].

Endocrinology assessment was done to check the blood sugar level 0,9 g/L and HbA1c was 6,8%.

The patient received antibiotics (amoxicillin/clavulanic acid 3g/day and metronidazole 3g/day) and antalgics (paracetamol 3g/day) daily, in parenteral, for 10 days, with removal of the causal tooth.

A decision to refrain from excising the tumor was taken after a multidisciplinary consultation.

Postoperative periods were favorable with the disappearance of swelling and alleviation of symptoms.

The patient was followed up in our specialized consultation; any clinical signs that appeared were mentioned on the patient's discharge form. Routine follow-up 15 days, 1, 3, 6, and 12 months later showed no signs of recurrence of symptoms like inflammation or discoloration of the skin and mucous tissues, increase in jugal or lingual mass, pulsation in the mass, pus issue, hemorrhage.

3. Discussion

Arteriovenous malformation is a direct connection between an artery and vein without capillaries in-between. Arteriovenous malformations may be discovered at birth or in childhood, others may be late onset in adulthood [8,9].

Intracranial localization is the most frequently affected site in the head and neck. The tongue and striated muscles are an extremely rare localization [10,12].

AVMs are normaly composed of a central nidus with anomalous arteriovenous shunts and a network of surrounding collateral vessels [6]. The clinical aspect of the lesion, the anatomical change and its evolution over time is explained by the short circuit between high blood pressure and low venous pressure [11]. This pressure depends on several factors that have modified it, such as the infection, trauma, ligation, attempted excision, an ischemic event secondary to thrombosis or ectasia, hormonal changes such as pregnancy or puberty [7]. For our patient, cellulite from dental origin was a triggering factor in the growth of her MVA and its extension.

The causes of vascular tumors are unknown. The pathogenesis of AVMs is still unclear, endothelial dysfunction theory has raised recently due to evidence of endothelial-derived somatic mutations of mitogen-activated protein kinase 1 in AVM [5].

The diagnosis is usually made with Doppler ultrasonography. However, CT scans look for bone involvement and extension while MRI scans look for soft tissue involvement. CT angiography (CTA) and magnetic resonance angiography (MRA) are useful imaging studies [13]. Regardless, angiography remains the diagnostic gold standard and helps identify the central nidus and feeder arteries, information that can then be used for treatment planning and embolization [14].

Diffuse AVMs are rarely cured but easily controlled with current treatment options.

Common treatment for AVMs for extra cranial AVMs of the head and neck is embolization and surgery. Medical treatment still not yet been proven in the stabilization or removal of extra cranial lesions [15,16].

The embolization consists of: the direct injection of agents caustic to the vascular endothelium under radio-graphic guidance and arteriography. Instead, embolization alone had showed a high recurrence rate (98%) [17,18]. For best treatment results, embolization is followed by complete surgical resection 6–48 hours later. In some cases embolization prior to surgery can be misleading, and reduces the size of the mass to be excised, thus increasing the rate of recurrence [19].

The surgical treatment consists of an excision of the lesion which may be preceded by embolization with reconstruction of the loss of substance. The limits of exeresis remain an emblematic decision, for the

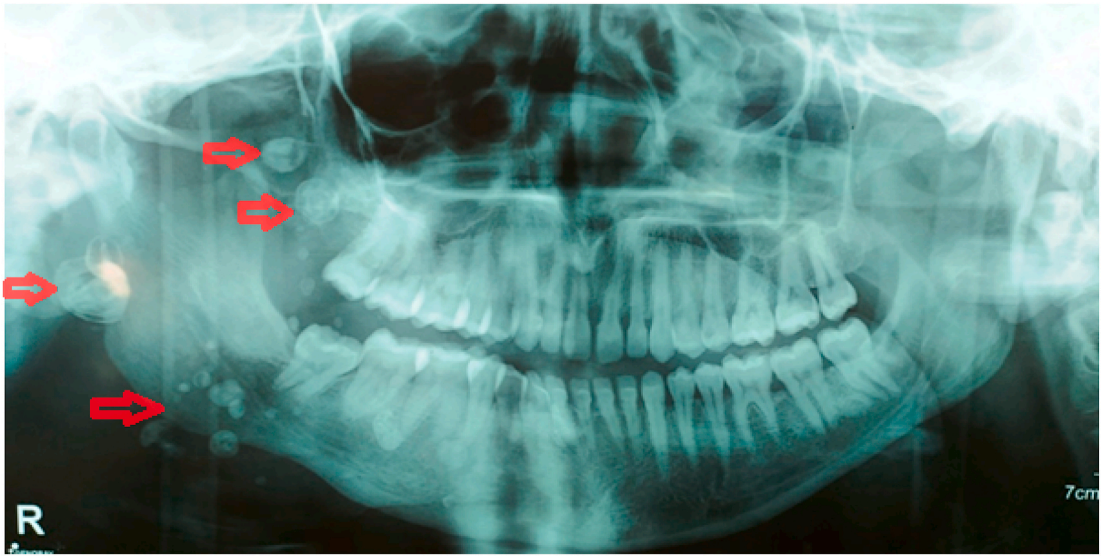

Fig. 3. Dental x-ray picture showing radio-opaque lesions in the right mandible.

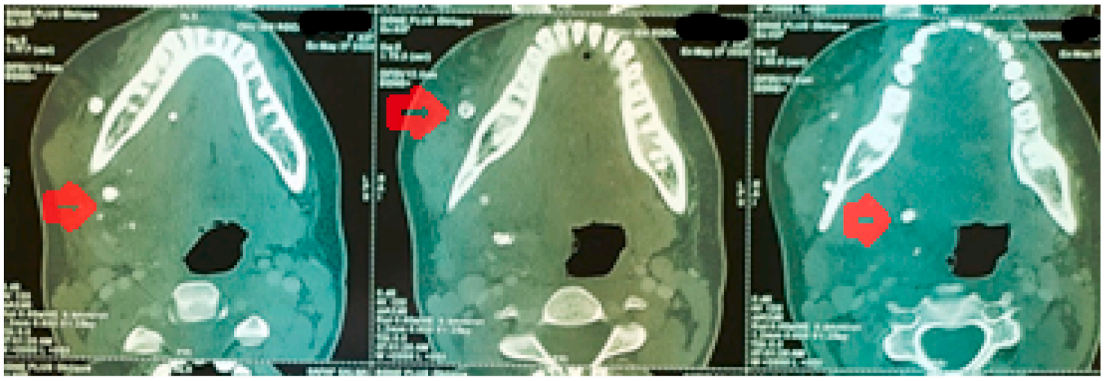

Fig. 4. Axial computed tomography picture showing phleboliths in the right jaw.

diffuse VMAs, it can be over-exerted or under-resected [18].

Several adjuvant treatments are used, such as laser therapy for small superficial vessels, and interstitial therapy by injecting a caustic agent into the endothelium of the vessels to reduce tissue burden in superficial disease [7].

Due to the high recurrence rate and the severity that the disease can take, patient monitoring and follow-up must be frequent. the request for paraclinical examinations is necessary and depends on the clinical condition and progression [7].

Concerning our observation, it is a massive arteriovenous malformation evolving since childhood; complicated by cervicofacial cellulitis. After a multidisciplinary consultation, we advocated close monitoring instead of mutilating removal. Regarding the evolution and prognosis, the follow-up was simple after parenteral medical treatment and facial massage. At 10 days, there was an improvement of the mouth opening with disappearance of the pus issue, Currently the mass has regressed in size.

4. Conclusion

The management of head and neck AVMs requires multidisciplinary and multimodal care by appropriately trained physicians.

The rarity and unpredictable evolution of this type of tumor makes its management quite difficult, a commitment of the patient and multidisciplinary doctors is necessary.

It is necessary to treat localized AVMs quickly and to be very careful in the treatment of diffuse AVMs, so as not to wake up the volcano and have a more extensive and aggressive recurrence, and above all to inform the patient to avoid aggravating factors.

Patient consent

The consent to publish this information was obtained from study participant. We confirm that written proof of consent to publish study participant are avaible when requested and at any time.

Consent and ethical approval

As per university standard guideline, participant consent and ethical approval have been collected and preserved by the authors.

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Author contribution

Amine kaouani: Corresponding author writing the paper.
Ouassime kerdoud: writing the paper.
Rachid Aloua: writing the paper.
Faiçal Slimani: Correction of the paper.

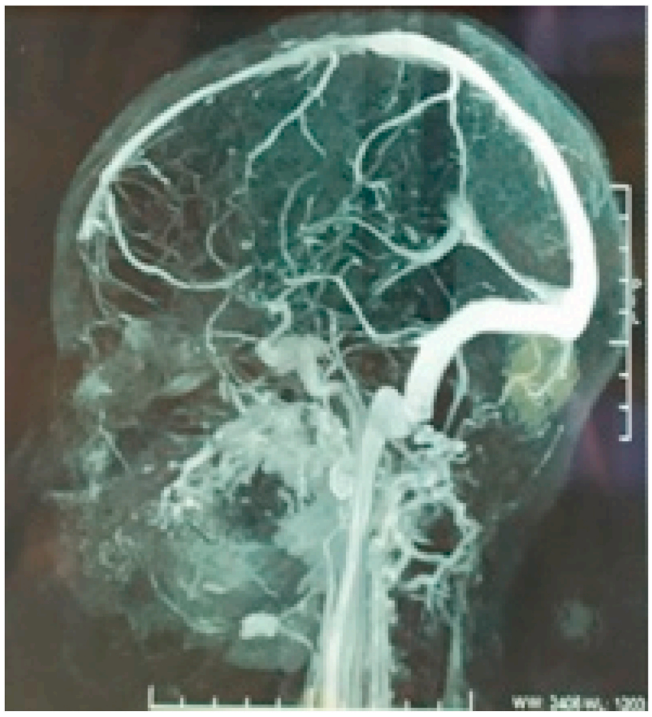

Fig. 5. Arteriography shown a diffuse AVMs.

Guarantor

Amine kaouani.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

Authors of this article have no conflict or competing interests. All of the authors approved the final version of the manuscript.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102196.

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