

## ORIGINAL ARTICLE

# Arteriovenous Malformation of the Buccal Mucosa: A Rare Case Report

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### ABSTRACT

*Introduction: Arteriovenous malformations are high-flow lesions that result from persistent direct arterial and venous communication. Although they are present from birth, they may not become noticeable until later in childhood or adulthood. This is a rare lesion and their preferable location in the body is not known exactly. The aim of this study was to report an arteriovenous malformation in the buccal mucosa. Materials and Methods: A 45 years old male referred to private office. A mass with soft consistency was observed on the clinical examination. With clinical impression of lipoma, surgical resection was done. H&E serial sections revealed a mixture of thick-walled arteries and veins, along with capillary vessels in the background of lipomatous connective tissue and pathology diagnosis was reported as benign vascular lesion compatible with arteriovenous malformation. Discussion & Conclusion: After observing the resected specimen under light microscopy and consultation with the surgeon, He reported intensive hemorrhage during surgery. Because of lack of any case report in the literature at the time of we diagnosed this lesion, it was so difficult to sign the report as an arteriovenous malformations of the buccal mucosa especially with the lack of angiography. Fortunately, surgical procedure information, given by the surgeon was so helpful in early diagnosis and confirmation of histopathologic findings. Our regular follows up showed no recurrence after 4 years monitoring of the patient. The close relationship between clinician and pathologist and importance of sending all clinical and Paraclinical data to laboratory are strongly emphasized here.*

**Keywords;** Arteriovenous malformation, Arteriovenous hemangioma, Buccal mucosa, Oral mucosa, Oral cavity, AVM

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### INTRODUCTION

Arteriovenous malformations are high-flow lesions that result from persistent direct arterial and venous communication. Although they are present from birth, they may not become noticeable until later in childhood or adulthood. Because of the fast vascular flow through these lesions, a palpable thrill or bruit often is noticeable. The overlying skin typically feels warmer to touch. Presenting symptoms may include pain, bleeding, and skin ulceration [1].

AVM might be found in various sites in the head and neck. Most of the reported cases have been occurred within the midface and mandible [2-5]. We found just a new case of AVM in the literature reported recently by Stephanie T, in the buccal mucosa of a 76 years- old female [6]. However, it can be difficult to distinguish AVM from various soft tissue tumors and vascular anomalies of the buccal mucosa [7-8]. Because of the progressive and destructive enlargement of AVMs along with their inherent surgical risks, accurate diagnoses of AVMs is critical.

Unfortunately, there is not established consensus on treatment of AVM. The treatment could be surgery (enucleation or resection) or interventional radiology (arterial embolization or transosseous transcatheter embolization). There are several indications for treatment, including age, and the size and type of vascular malformation (active or passive) [9].

Hemangiomas and vascular malformations have been recognized as distinct diseases that exhibit unique properties and behavior that demand an appropriately tailored treatment plan. Different treatment modalities are advocated for different lesions: injection of平阳霉素 for venous malformations, Krypton laser photodynamic therapy of venular malformations, Nd:YAG laser therapy for deep head and neck venous malformations after surgical exposure of the lesions, as well as "double" embolization of large venous malformations.<sup>10</sup>

According to the clinical characteristics, patients with maxillofacial AVMs could be divided into the 4 types: cystic dilatation lesions, limited thicken lesions, diffuse thicken lesions, central maxillary hemangioma. This new type of clinical classification is beneficial for selecting method of treatment (operation, embolization of supplying artery alone, embolization of supplying artery plus hardener intratumorous injection or embolization of supplying artery plus tumor resection). Preparing a good digital subtraction angiography is necessary for maxillofacial AVMs. The embolization of tumor supplying artery alone could cure the small AVM with single branch terminal blood supply. The embolization of supplying artery plus hardener intratumorous injection or the embolization of supplying artery plus tumor resection is an effective method for maxillofacial AVMs.<sup>11</sup>

### CASE REPORT

A 45 years old male referred to private office. Facial deformity is obvious in the figure 1A. A soft mass with enlargement, pulsations and thrill was observed on the clinical examination. To rule out soft tissue tumors, surgical resection was done (Fig. 2A, B).



**Fig1. Facial deformity is seen in the right side (A). Facial deformity is not seen after 6 month follow up(B).**



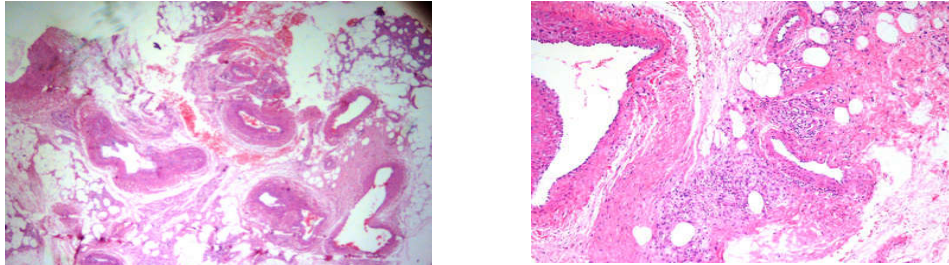
**Fig2. The large mass is obvious during surgery (A). Grossly, the red mass measuring 8X 2.5X 1.5cm, with soft consistency and solid cut surface is seen (B).**

The lesion could be categorized in stage 2 according to the Schobinger clinical staging system for AVMs, introduced at the 1990 meeting of international Workshop for the Study of Vascular Anomalies. This system is useful for documenting the presentation and evolution of the lesion. Stage 1 (quiescence) is characterized by a cutaneous blush, warmth, and arteriovenous shunting by Doppler examination. Stage2 (expansion) is similar but with enlargement, pulsations, bruit, thrill, and tense and/or tortuous veins. Stage3 exhibits dystrophic skin changes, ulceration, tissue necrosis, bleeding, and persistent pain. Cardiac failure constitutes progression to stage 4.<sup>12</sup>

According to the oral and maxillofacial surgeon the bleeding control was incredibly difficult during the surgery. The sample was sent to our laboratory with clinical impression of hemangioma and angioliopoma. Surprisingly, we found a few reports on the frequency of vascular malformation in Iran which stated that the frequency of hemangioma (including vascular malformations) vary from 0.6% to 3.5%. The absence of

enough information about the frequency of different kind of vascular malformation might influence on clinicians' point of view to make proper differential diagnosis. 13-16. No radiography or angiography had been performed before surgery. After surgical resection, specimen was sent to Pathology Laboratory, processed and stained by H&E.

Histopathologic findings: Serial sections revealed a mixture of thick-walled arteries and veins, along with capillary vessels in the background of lipomatous connective tissue (Fig3A, B).



**Fig3. H&E sections: look at thick-walled arteries and veins in the lipomatous background (A. ×100). The thick-walled vein (left) and capillary vessels (right center) are seen in B (×400)**

## DISCUSSION

Features of AVMs can be detected both radiographically and histologically. Then the distinction of AVMs from other vascular anomalies is allowed when their diagnosis is clinically (only by history and physical examination) elusive. Unfortunately the classic features of AVMs including palpable pulsations, spongy texture, and warmth are not always present. Salient features include deep, compressible soft tissue mass with brisk vascular rebound and an overlying mucosal or skin blush. Patients may report a focus of hyperemia at birth that has progressed to the current lesion. Trauma, hormonal changes, spontaneous internal bleeding, and prior interventions have also been associated with sudden progression and subsequent recognition of the disease. 17, 18

Clinical diagnosis should be confirmed by US and color Doppler examination. Angiography shows variable degrees of arterial dilatation and tortuosity, arteriovenous shunting, and dilated draining veins. Angiography is rarely performed unless intervention is planned under the same anesthetic. 12 Unfortunately in our presented case no angiography or color Doppler examination was requested by physician. If the surgeon used preoperative procedures, the patient management during surgery could be easier and more rationale. Clinicians should be aware of the potentially life threatening complications of such lesions, especially when surgery is performed without preparing angiography or other complementary techniques.

In our study, the above histopathology findings, along with clinical data, confirmed the diagnosis of arteriovenous hemangioma or the recent recommended name "arteriovenous malformation" (AVM). As in other head and neck AVMs, urgent intervention is warranted, especially when the lesion becomes clinically apparent. Researches show early diagnosis and surgical intervention improves the likelihood of cure while reducing undesired cosmetic and functional outcomes in young patients.

In our case, it seems that the surgical resection was curative, because there is no recurrence after 1 year follow up and the patient is satisfied because no deformity is seen in his face (compare Fig.1A and Fig.1B). Regular annual follow up was recommended for this patient, to monitor any signs of recurrence. Fortunately no recurrence has been observed after 4 years follows up.

Finally we strongly recommend the close relation between clinician and pathologist. It could be helpful especially when we encounter an uncommon lesion or we do not have all Para clinical information.

## CONCLUSION

This case demonstrates an AVM of the buccal mucosa rarely reported in the literature. The patient was successfully treated by surgical excision without recurrence after 4 years annual follows up. Since many clinicians may not be aware of the potentially life threatening complications of AVM, it is so important to be detected and treated early. Paraclinical examinations like Ultra Sound or color Doppler examination could be helpful for clinical diagnosis. For monitoring recurrences, regular follow up are strongly recommended.

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