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Central / Peripheral Vascular Malformationincranio-Facial Region Accidentally Discovered. (Case Report)

Tawfeeq BA[1], Al-Rawee RY [1*], Mohammed S[2], Saeed SS[3] and Mohammed SB[1]

[1]Department of Oral and Maxillofacial Surgery, Al-Salam Teaching Hospital, Mosul

[2]Department of Cardio-Vascular Surgery, Al-Salam Teaching Hospital, Mosul

[3]Department of Anesthesia, Al-Salam Teaching Hospital, Mosul

***Corresponding author:**

Rawaa Y Al-Rawee,
Department of Oral and Maxillofacial Surgery,
Al-Salam Teaching Hospital, Mosul, Iraq,
E-mail: dr.rawarawi@yahoo.com

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1. Abstract

1.1. Objectives: The need for direct supportive decision for management of central Vascular Malformation (VM) in emergency and elective conditions. Case Presentation: A 19-year-old male patient was referred to the MaxilloFacial clinic with massive oral bleeding from the mucosal sulcus of the left side of the cheek at the level of upper first molar tooth following accidental trial of incision and drainage of suspected abscess in private clinic, the case managed in casualty unit and operated later on as a partial maxillectomy with ligature of facial artery.

1.2. Discussion: We present a very rare case with characteristics of central and peripheral VM, including the patient's: clinical history; findings of examination; and radiographic and scanning examination.

2. Introduction

Hemangiomas are benign vascular neoplasm's characterized by an abnormal proliferation of blood vessels [1]. Cavernous Hemangiomas are benign proliferations of vessels that usually occur at or just after birth [2]. Intra-osseous Arterio-Venous Malformation (AVM) in the craniofacial region is rare [3]. Head trauma may have be the cause, localized headache can be seen with this pathology [4]. Intra-osseous vascular lesions are subdivided into hemangiomas and Vascular Malformations(VM) [5]. Its early detection and treatment are important as any minor trauma may result in fatal hemorrhage [6].

Hemangiomas and VM can cause significant morbidity and even mortality. Physicians often confuse these lesions. The nomenclature for classifying these lesions is often used interchangeably and inappropriately. Clinically significant malformations are uncommon. Both lesions are endothelial malformations [7]. VM are always present at birth and enlarge with growth and do not in volute and remain present throughout the patient's life [8].

Historically, lesions were named according to the size of the channels and the type of fluid content. Blood-containing lesions were called hemangiomas and were separated into capillary, strawberry, and cavernous on the basis of channel size. Lymph-containing lesions were referred to as lymphangiomas or cystic hygromas [7]. This classification system replaced by one described in 1982 by Mulliken and Glowacki [8]. Which separates endothelial malformations into hemangiomas and VM, on the basis of their natural history, cellular turnover, and histology [8]. A more pertinent issue is classifying vascular malformations as either low-flow or high-flow lesions [9]. Surgical excision combined with intravascular embolization is treatment of choice for AVM with the knowledge that extensive AVM are still not curable [10]. Irradiation is suggested if the lesion is large or surgically inaccessible [11] with side effects such as damage to growth centers in developing patients and induction of neoplastic transformation [12]. Cryo-therapy have some measure of success in small lesions [13]. Embolization of large feeder vessels is also recommended under fluoroscopic

control with risk of pulmonary or cerebralembolization [14]. Embolization has been shown to be temporary [12]. Therefore, its primary use is adjunctive to surgery to reduce intraoperative bleeding [13]. Surgical intervention is generally accepted as the definitive treatment, with en bloc resection the recommended procedure [15]. Ligation of feeder vessels should precede the removal of the lesion [9].

3. Case Presentation

3.1. History: A 19-year-old male patient with a fully dentate maxilla was referred (27 Nov. 2013) to the clinic of Department of Oral and Maxillofacial Surgery, Al-Salam Hosp., Mosul. The lesion focused to the mucosal sulcus of the left side of the cheek at the level of upper first molar tooth, the site show a profuse bleeding following accidental trial of incision and drainage of suspected abscess in private clinic, the bleeding was massive that cannot be controlled even with the local power full suction machine, we try to suture the traumatic defect under local anesthesia with pressure pack intra- and extra-orally secured with Barton bandage.

3.2. Clinical Finding: In the clinical examination, there is no any evidence in the face that may indicate the vascular lesion on the skin surface, even simple swelling, the same is noted in oral examination except mild buccal posterior mucosal bulge in the upper left side. Patient had a full dentated upper and lower jaw, with sound immobile standing teeth in the affected site. Normal mucosal color overall the mouth.

Patient admitted to our ward, with careful follow up and hematological investigation reveal absence of bleeding disorder, with proper general health, patient had negative history regarding any clinical signs or symptoms related to the present chief complain, radio graphical view reveal normal maxilla with small radiolucenyin the area between the apices upper left lateral incisor and canine, looks unrelated to the location of the relevant problem, CT scan and MRI reveal normal radio graphical report without any abnormal finding, after 6 days of hospital admission, the patient dismissed home with healed suture site.

One day later, he admitted to the casualty unit following massive uncontrolled osseous bleeding at the area above upper left lateral incisor and canine, with failed multiple local trials to control the massive bleeding, finally, the space in the upper jaw created by extraction of both upper left lateral incisor and canine and destruction of the inter-septal bone with pressure gauze inserted in the bony gap, additional gauze overlying the deep one, and instruct the patient to bite over the bulky gauze, at that time, the patient pass clinically to hypovolemic shock, which opposed immediately with fast forcible transfusion of two pints of blood, the case discussed in temporary multi-disciplinary specialty team (vascular surgeon, hematologist, radiologist, anesthetist),then the patient prepared for surgical work under general anesthesia in association with cardiovascular specialist two days later.

3.3. Imaging Findings: Periapical view (accompanied with the patient) reveals a small radiolucent area located in between the apices upper left lateral incisor and canine measure about (4x6) mm. Angio CT of the external carotid artery (left side) was performed, with an evidence of expansible lesion seen involving the left side of the maxillary region, involvement of the alveolar margin, with loss of related teeth. The lesion reveals 40-60 HU density with inner and external components. Prominent left facial artery seen. The change noted could be consistent with intra – osseous maxillary vascular anomaly. Hematological tests reveal normal blood film with absence of bleeding disorders.

3.4. Surgical Management: The patient prepared for surgical work in the theatre, the vascular specialist temporarily ligate the external carotid artery and clinically enlarged facial artery, we start with planed partial maxillectomy, starting with extraction of left central incisor which results in profuse bleeding, then extract right central incisor and left first premolar, reflecting buccal and palatal flaps, using round drill on surgical hand piece to delineate the borders of the partial maxillectomy, the osteomised segment separated with osteotome and surgical mallet, the massive bleeding declines prominently, the exposed cancellous bone coagulated using electrical diathermy, sealed with bone wax packed with softened impression compound, the buccal mucosa posterior to the maxillectomy site (the site of peripheral VM) ligated externally with 1:0 round silk suture to control the soft tissue excessive ooze, finally, the temporary carotid ligature released while the facial artery occluded permanently (Figure 1).

Patients pass the post-operative period safely with only one pint matched blood. The patient evaluated two days later, the temporary splint and bone wax removed with insignificant bleeding, packed and carefully followed for one week in the ward, then dismissed home with periodic weekly follow up.

For the soft tissue VM, the decision was to be kept under observation for conservative treatment with periodic follow up, or otherwise selective angiographic therapy.

Histopathology: the histopathological finding of the excised bony biopsy (4cm, 2.5cm, 3cm) reveal loose fibrovascular tissue with small thick arteriols and venous channels with capillaries, foamy cells indicating arterio-venous malformation.

3.5. Doppler Finding:

- 10 days post-operative, US of the left Maxilla, poorly defined isoechoic mass (6x2)cm involving deep and superficial layers of buccinators muscle, extend to periphery of left upper lip and lateral to the left nasal base subcutaneous tissue (along distribution of facial artery), suggestive of widely distributed hemangioma. CDUS reveals heavy vascular flow within the mass with widen caliber of its feeding artery about 4.5 cm.

- Three months postoperative, still, poorly defined isoechoic mass like lesion (34x12)mm contains no calcification, suggestive of resolving hemangioma or AVM. CDUS reveals few vascular pedicles within this mass looks related to left facial artery.
- Four months postoperative, ill-defined mass about (30x9) mm seen at left maxillary area along the course of facial artery. CDUS show few vascular signals inside, no calcification.

In convenience with all previous diagnostic measures, the case is diagnosed as fast-flow bony and soft tissue vascular malformation, regarding the last Doppler reading, the decision is to keep the patient on a conservative periodic follow up, for one visit monthly to check the progression of the presenting condition. For the created surgical defect in the jaw, the patient refuses any prosthetic replacement.

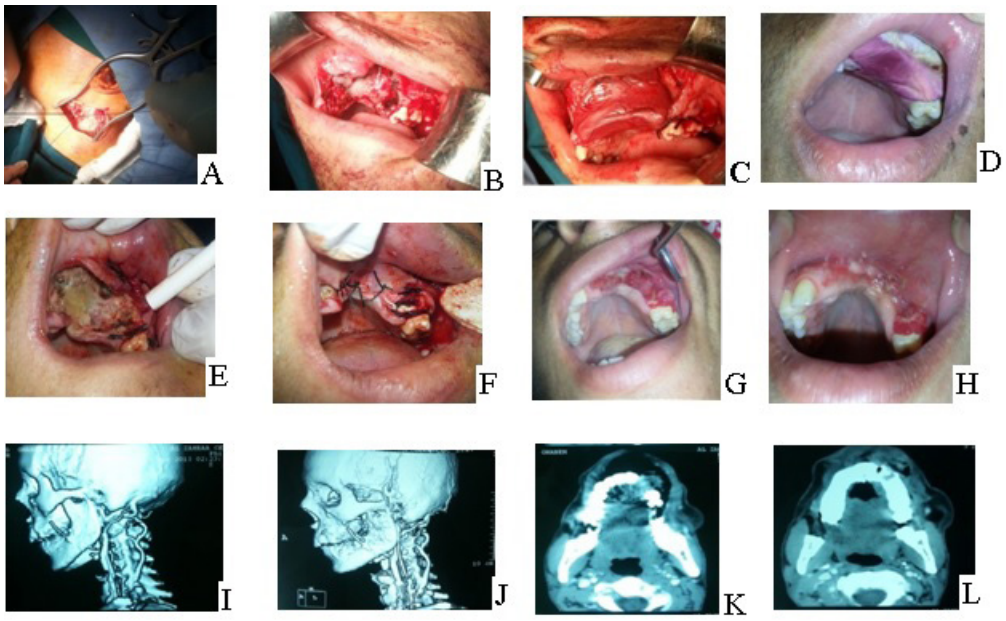

Figure1: Serial pictures for the case,
A: Temporary ligation of external and facial arteries prior to partial maxillectomy.
B-C: Intra-operative partial maxillectomy site and placement of impression compound material.
D,E, F: Two days postoperative, removal of the compound pack and bone wax and placement of iodofoam pack with mucocal suture.
G,H: one and three month's postoperative maxillary defect following partial maxillectomy.
I-L: Angiography CT Scan reveal left facial artery enlargement with central VM.

4. Discussion

In this present study; authors emphasize a very rare case illustrates many features that are characteristic of fast-flow central and peripheral VM, including the patient's clinical history; findings of examination; and radiographic and scanning examination. Emergency bleeding situation should be controlled and managed with accepted degree of additional faults such as teeth loss or bone destruction or work without local anesthesia, and sometimes without some rules and principles keeping in mind saving patient life.

Radiotherapy was not the treatment of choice, considering the needs for emergent management, age of the patient and the retarding effects of radiation on oral and perioral tissues. Intra-lesional injections of sclerosing agents would not have been effective because of the lesion's bony nature, this is in association with Nikhil Marwah et al [15]. Hence, in this case, surgical partial maxillary resection was the treatment of choice due to multiple factors such as the: lesion size; potential active bleeding; and lesion's approachability. The maxilla was resected beyond the lesion's radiographic boundaries to avoid any manipulation of the vascular lesion and to prevent any complications, such as extensive hemorrhage [16].

Because of the serious consequences, VM must always be considered in the differential diagnosis and proper precautions must be taken in establishing the final diagnosis before any surgical treatment is undertaken.

In a recent study published by Jeong Woo Lee and Ho Yun Chungin 2020; they claimed that "AVM can cause disfigurement, compression, or destruction of adjacent tissues. Although AVM is considered a quiescent lesion, angiogenesis (growth of new blood vessels

from preexisting vasculature) and/or vasculogenesis (de novo formation of new vasculature) may be involved in AVM expansion". As well as the stated that treatment protocol is to control AVM whether superficial or deep in a way of different interventions can include embolization, resection, or a combination is focused on reducing symptoms, preserving vital functions, and improving deformities. This was gain in this case report management [17].

Finally; It's important to highlight an essential points have been discussed scientifically in clear manner by Xiao Li and his colleagues in published article 2020; such as AVM consider as one of the most dangerous haemorrhagic diseases in maxillofacial region with a high tendency of life-threatening haemorrhage. AVM occurred in the mandible more often than in the maxilla (64.93% and 32.23%, respectively) as well as 95.26% of cases occurred in the posterior teeth region [18].

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Consent:

Written informed consent was obtained from the guardian of the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal if need.

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