ARTERIOVENOUS MALFORMATIONS IN THE DIFFERENTIAL DIAGNOSIS OF PALATAL SWELLINGS*

Palatal Şişliklerin Ayırıcı Tanısında Arteriovenöz Malformasyonlar

Bora ÖZDEN1, Burcu BAŞ1, Hatice DURAN1, Peruze ÇELENK2, Ömer GÜN Han3

Received: 14/01/2014
Accepted: 01/03/2015

ABSTRACT

An arteriovenous malformation (AVM) is composed of abnormal communications between arteries and veins without the normal intervening capillary bed. AVM of the head and neck is a rare vascular anomaly. We present here an unusual case of AVM with the size of 4x3 cm at the left posterior palatal area. Incisional biopsy revealed AVM. Resection of the lesion following angiography was suggested to the patient however, he refused the treatment. The patient was considered to be under control. AVM should always be kept in mind in the differential diagnosis of palatal swellings.

Keywords: arteriovenous malformation; palatal swellings

ÖZ

Arteriovenöz malformasyon (AVM), arterler ve venler arasında normal kapiller yatak olmadan anormal bağlantı ile oluşmaktadır. Baş ve boyunda görülen AVM nadir görülen vasküler anomalidir. Sol posterior palatinal bölgede 4x3 cm boyutlarında bir AVM olgusu sınındaktayız. İnsizyonel biyopsi sonucu AVM olarak tespit edildi. Hastaya anjiyografi takiben lezyon rezeksyonu önerildi fakat hasta tedaviyi reddetti. Hasta kontrol altına alındı. AVM palatal şişliklerin ayırıcı tanısında her zaman akılda tutulmalıdır.

Keywords: arteriovenous malformation; palatal swellings

Anahtar kelimeler: arteriovenöz malformasyon; palatal şişlikler

1 Department of Oral and Maxillofacial Surgery Faculty of Dentistry Ondokuz Mayıs University
2 Department of Oral and Maxillofacial Radiology Faculty of Dentistry Ondokuz Mayıs University
3 Department of Oral Pathology Gülhane Military Academy

*This article has been presented as a poster in 6th ACBID International Oral & Maxillofacial Surgery Society Congress" May 26-30, 2012, Antalya, Turkey.

This work is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License.
Arteriovenous malformation of the palate

Introduction

Arteriovenous malformations (AVM) are rare lesions which can easily be misdiagnosed yet produce the very dramatic clinical presentation of severe life threatening oral bleeding. They are composed of abnormal communications between arteries and veins without the normal intervening capillary bed (1-3). Blood from the normal circulation increases the size of such a malformation through a low resistance system created by the nidus, the aberrant arteriovenous network at its centre (1, 4). Their real importance lies in their potential to result in exsanguination which usually followed by an unrelated treatment, such as tooth extraction, surgical intervention, puncture wound or blunt injury in the involved areas, without awareness of the existence of the AVM.

AVM can occur as an acquired event, especially following trauma, but usually is congenital (3). Trauma, an ischemic event secondary to thrombosis, ectasia, hormonal changes and puberty can induce proliferation of the AVM (5). Bone involvement occurs in 35% of cases (5-7). Vascular lesions are seen as often as 50% in the head and neck region (8). Although they are often found in the head and neck, they rarely exist in the oral cavity. Treatment of these lesions are complex, and decision of the therapies involve balancing the risks and benefits of various treatment options such as surgical excision, embolization, radiosurgery, and conservative management (9). Central arteriovenous malformations of jaws, although rare, are clinically important owing to the potential risk of life-threatening hemorrhage.

The aim of this report is to present the diagnosis of a rare case of AVM at the maxillary palatal region.

Case Report

A 58 year old male patient was referred to our department with a 20 years’ history of a painless swelling at the left palatal area. A doughy mass about 4x3 cm in diameter was found at the posterior palate on clinical examination (Figure 1). The entire area appeared relatively normal at orthopanthomograph (Figure 2).

T1-weighted MRI scans showed a mass located in the left hard palate (Figure 3). No fluid was obtained on aspiration with a large-bone needle. Considering the localization and clinical view of the lesion, pleomorphic adenoma was thought for the initial diagnosis. The incisional biopsy was performed on two sections. During the biopsy, a thick layer of fat tissue and submucosa was observed. Histopathological examination showed a mesenchimal neoplasm with numerous small blood vessels, the vasculature was embedded in a myxoid matrix (Figure 4). Finally the lesion was evaluated as AVM. Resection of the lesion following angiography was suggested to the patient but he refused treatment so the patient was considered to be under control.

Figure 1. Palatal swelling at the left side of the palate.

Figure 2. There was no evidence of the lesion in the panaromic radiography.
Discussion

The differential diagnosis for palatal swellings includes reactive lesions (e.g., fibroma or fibroed pyogenic granuloma, or lymphoid hyperplasia), palatal abscess, salivary gland neoplasms, and malignant tumors (10). In our cases, the possibility of palatal abscess was ruled out due to absence of signs of inflammation. No etiologic irritation factor was detected for fibrous lesions. The lack of ulceration of the palatual mucosa or invasion of the surrounding tissue rules out the possibility of malignant transformation. We also ruled out hemangiomas though they are rarely found on the hard palate and often seen as red-bluish in color. In our cases the mucosa was in normal color and appearance. We clinically diagnosed the lesion as pleomorphic adenoma of the minor salivary gland due to its localization and clinical appearance. However, histopathological examination after the incisional biopsy was revealed as AVM. As a result, we emphasize the importance of incisional biopsy before any surgical treatment of the lesions of hard palate. “Vascular malformation” is a generalized term used to describe a group of lesions, present at birth, formed by an anomaly of angiovascular or lymphovascular structures. Vascular malformations occur in approximately 1% of births but majority of these patients do not present for treatment (11). The high-flow vascular anomalies in the head and neck are arteriovenous malformations (AVMs) (12). These are the lesions with direct communications between an artery (or arteries) and a vein (or veins) by passing the capillary bed (13). Almost all patients presenting with AVM are children or adolescents (14-16). However, in this case, 58 years old patient was presented. In the oral cavity, these can present at any site, but most commonly on anterior two-thirds of tongue, leading to macroglossia and difficulty in mastication, speech, and deglutition. Other sites that may be involved are palate, gingiva, and buccal mucosa (17). Angiography is currently the gold standard for determination of location and flow characteristics of vascular lesions (18). Angiography can differentiate between the different types of vascular lesions and can help provide visualization in real time for embolization. With angiography, it is possible to determine which blood vessels are supplying the lesion, and the relative venous outflow characteristics and presence or absence of arterio-venous shunts, which are important in determining the appropriate embolization techniques to employ (19). Ligation of the external carotid artery (ECA) is completely proscribed, because even if it stops hemorrhage it never prevents recurrence from the rich collaterals and therefore it increases the difficulty of further treatment. Complete resection of the involved bone may be curative, but it involves severe blood loss and induces damage to the shape and function of the involved bone. Radiotherapy often fails owing to the level of cellular maturation along with the danger of radiation damage. Complete cure by arterial embolization is difficult, either with particles or glue, and may involve potential complications. Conservative resection after arterial embolization could be safe and curative, but it still involves functional deficits (20, 21). In this case; we suggested resection of the lesion following angiography to the patient but he refused treatment so the patient was considered to be under control.

Conclusion

AVM should always be kept in mind in the differential diagnosis of palatal swellings. Both the radiologist and
the surgeon need to be aware of its diverse presentation as it may influence treatment protocol.

**Source of funding**
None declared

**Conflict of interest**
None declared

**References**

1. Buckmiller LM, Richter GT, Suen JY. Diagnosis and management of hemangiomas and vascular malformations of the head and neck. Oral Dis 2010;16(5):405-418.
2. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: A classification based on endothelial characteristics. Plast Reconstr Surg 1982;69(3):412-422.
3. Persky MS. Congenital vascular lesions of the head and neck. Laryngoscope 1986;96(9 Pt 1):1002-1015.
4. Richter GT, Suen J, North PE, James CA, Waner M, Buckmiller LM. Arteriovenous malformations of the tongue: A spectrum of disease. Laryngoscope 2007;117(2):328-335.
5. Nocini PF, Fior A, Tolo C, Bertossi D. Arteriovenous malformation of the nasal ala: A case report. J Oral Maxillofac Surg 2000;58(11):1303-1309.
6. Boyd JB, Mulliken JB, Kahan LB, Upton J, 3rd, Murray JE. Skeletal changes associated with vascular malformations. Plast Reconstr Surg 1984;74(6):789-797.
7. Sadowsky D, Rosenberg RD, Kaufman J, Levine BC, Friedman JM. Central hemangioma of the mandible. Literature review, case report, and discussion. Oral Surg Oral Med Oral Pathol 1981;52(5):471-477.
8. Kennedy KS. Arteriovenous malformation of the maxilla. Head Neck 1990;12(6):512-515.
9. Marshall GA, Jonker BP, Morgan MK, Taylor AJ. Prospective study of neuropsychological and psychosocial outcome following surgical excision of intracerebral arteriovenous malformations. J Clin Neurosci 2003;10(1):42-47.
10. Scheper MA, Nikitakis NG, Meiller TF. A stable swelling of the hard palate. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104(4):461-464.
11. Martines F, Immordino V. Arteriovenous malformation of the base of tongue in pregnancy: Case report. Acta Otorhinolaryngol Ital 2009;29(5):274-278.
12. Chiu YW WH, Chan YW, Lui MT, Kao SY, Lo WL. A giant venous malformation of face and neck: A case report. Taiwan J Oral Maxillofac Surg 2011;22:110-117.
13. Duncan IC FP. Vascular malformations. Part 2. Current classification of vascular malformations. South African J Radiology 2004;8(1):23-30.
14. Churojan A, Khumtong R, Songsaeng D, Chongkolwatana C, Suthipongchai S. Life-threatening arteriovenous malformation of the maxillomandibular region and treatment outcomes. Interv Neuroradiol 2012;18(1):49-59.
15. Sakkas N, Schramm A, Metzger MC, Berlis A, Schmelzeisen R, Otten JE, Hohlweg-Majert B. Arteriovenous malformation of the mandible: A life-threatening situation. Ann Hematol 2007;86(6):409-413.
16. Sasaki R, Okamoto T, Komiya C, Uchiyama H, Ando T, Ogiuchi H. Mandibular gingival arteriovenous malformation in pregnancy. Br J Oral Maxillofac Surg 2008;46(8):675-676.
17. Shetty DC, Urs AB, Rai HC, Ahuja N, Manchanda A. Case series on vascular malformation and their review with regard to terminology and catarization. Contemp Clin Dent 2010;1(4):259-262.
18. Jackson IT, Carreno R, Potparic Z, Hussain K. Hemangiomas, vascular malformations, and lymphovenous malformations: Classification and methods of treatment. Plast Reconstr Surg 1993;91(7):1216-1230.
19. Kademani D, Costello BJ, Ditty D, Quinn P. An alternative approach to maxillofacial arteriovenous malformations with transosseous direct puncture embolization. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;97(6):701-706.
20. Larsen PE, Peterson LJ. A systematic approach to management of high-flow vascular malformations of the mandible. J Oral Maxillofac Surg 1993;51(1):62-69.
21. Liu D, Ma X. Assessment of efficacy of endovascular embolization for central arteriovenous malformations (avm) in the jaw. Zhonghua Kou Qiang Yi Xue Za Zhi 2002;37(5):340-342.

**Corresponding Author:**
Hatice DURAN
Department of Oral and Maxillofacial Surgery
Faculty of Dentistry Ondokuz Mayis University
55139-Kurupelit-Samsun/TURKEY
Phone: +90 362 312 19 19/3017
e-mail: htcdrm84@yahoo.com.tr