International Journal of Oral & Maxillofacial Pathology. 2013;4(4):37-41
Available online at http://www.journalgateway.com or www.ijomp.org
ISSN 2231 – 2250

Case Report

Mixed Hemangioma of the Gingiva: A Case Report

Ahmed Chkoura, Wafae El wady, Bouchra Taleb

Abstract

Hemangioma is the most common benign oral soft tissue tumor in children. In adults, the occurrence of hemangioma with its primary location on gingival tissues is very rare. Two main forms of hemangiomas are recognized: capillary and cavernous. Surgical treatment of this lesion may produce a severe bleeding, for this reason; it may be managed by specialized practitioner. This article reports a case of hemangioma localized in the mandibular gingiva in a 19-year-old man, the adopted treatment was excision of the tumor with a narrow margin of normal mucosa. The histopathological examination shows that it consisted of a mixed capillary-cavernous hemangioma. The pathogenesis, differential diagnosis and the treatment of hemangioma of the oral cavity are discussed. According to our research, this is the first described mixed capillary-cavernous gingival hemangioma in the literature.

Key words: Capillary Hemangioma; Cavernous Hemangioma; Tumor, Gingiva; Oral Cavity; Mandible.

Ahmed Chkoura, Wafae El wady, Bouchra Taleb. Mixed Hemangioma of the Gingiva: A Case Report. International Journal of Oral & Maxillofacial Pathology; 2013:4(4):37-41.

Received on: 13/08/2013 Accepted on: 20/12/2013

Introduction

Hemangioma is a term that encompasses a heterogeneous group of clinical benign vascular lesions that have similar histologic features.[1] It is benign lesion, which is a proliferating mass of blood vessels and do not undergo malignant transformation. Although a few cases are congenital, most develop in childhood. Occasionally, older individuals are affected.[1,2] Although hemangioma is considered one of the most common soft tissue tumors of the head and neck; it is relatively rare in the oral cavity.[1] Hemangiomas are often classified on the basis of their histological appearance and two main types are described: capillary and cavernous.[3,4] Capillary hemangiomas are composed of many small capillaries lined by a single layer of endothelial cells supported in a connective tissue stroma of varying density. Cavernous hemangiomas are formed by large, thin-walled vessels or sinusoids lined with a single layer of endothelium which are separated by thin septa of connective tissues. Mixed hemangiomas consisting of both components may also occur.[4] The purpose of this paper was to report the case of a mixed hemangioma with a primary localization in the gingiva.

Case Report

A 19-year-old Moroccan male was referred by his dentist to our oral surgery department for evaluation and treatment of the gingival bleeding and overgrowth between the first and the second right mandibular molar that had started one year ago, according to the patient, his medical and dental history was unremarkable, and he did not take any medications. The patient consulted a dentist three months ago who performed the excision of the lesion which was incomplete according to the patient.

The remaining mass has been increased gradually in size since that time. Then the patient was referred by his dentist to our department. During the physical examination, no other similar lesions were clinically visible in the head and neck region. Moreover, no lymph nodes were palpable. Clinical evaluation revealed a mass on the buccal surface of lower right molar region (Fig 1a). It was firm, non-painful, pedunculated, attached to the interdental papilla. The mass bled easily upon palpation, but did not blanch. There was a mild accumulation of dental plaque and supragingival calculus around his teeth. Differential diagnoses of pyogenic granuloma and hemangioma were given. Radiographic examination revealed a faint radiographic shadow of the crestal bone between the first and the second right mandibular molar (Fig 1b). A root scaling was done to remove any local irritating factors that may have been responsible for

 ISSN 2231 - 2250

the gingival inflammation. The patient was educated regarding good oral hygiene maintenance practices. The patient was called two weeks later and the swelling persisted with no change in size or in coloration.

Excisional biopsy was then performed to remove the swelling and to confirm the diagnosis (Fig 1c & 1d). It produced profuse hemorrhage and was controlled by pressure with gauze. The lesion was removed with a narrow margin of normal mucosa. Resorption of bone on the buccal surface mesial root of the second molar and on the distal root of first molar was noted. Finally a careful curettage of the bone with periapical curettes was performed in order to ensure total removal of the lesion and to prevent recurrence. Microscopic examination of the excised tissue revealed a proliferation of vascular structures with different size varying from small capillaries to large vessels, lined with regular endothelial cells supported by thin connective tissue (Fig 1e). The diagnosis of mixed capillary-cavernous hemangioma of the gingival was posed. During the follow-up after three and six months later, the patient was free of recurrence but the buccal surface of the root of first and second mandibular teeth are still exposed, a periodontal surgery was planned to cover the defect.

Discussion

Hemangiomas are tumor-like malformations composed of seemingly disorganized masses of endothelium-lined vessels that are filled with blood and connected to the main blood vascular system.[5] The majority of hemangiomas involve the head and neck region, and more females are affected.[3,4] Retrospective studies of oral tumours in paediatric patients have shown that hemangiomas were the most common benign soft tissue tumour in children.[6-8] In a survey where a broad spectrum of biopsied oral lesions were studied, haemangioma accounted for 4.4% of all oral biopsies and was the sixth most common oral lesions in patients up to 15 years of age.[9] This was in contrast to earlier studies which found less than 1% of hemangiomas in oral biopsies from patients up to 19 years of age.[10,11] More recently Wang et al. have reported that haemangioma was the third most common pediatric benign oral tumor after the odontome and ameloblastoma.[12] However, all these figures are probably under-estimations of the true prevalence of hemangiomas in children as some of the lesions may have been left for natural involution and were not available for biopsy.[13] These lesions are mostly seen on the face, fingers and occasionally seen on oral mucosa.[14] They may occur as isolated lesions in the oral cavity, as multiple lesions affecting different parts of the body and in association with other developmental anomalies in the various angiomatous syndromes.[5] The occurrence of hemangioma with its primary location on gingival tissues seems to be extremely rare.[14]

Clinically hemangiomas of the oral soft tissues often appear as a soft mass, smooth or lobulated, sessile or pedunculated and may be seen in any size from a few millimeters to several centimeters.[14] They are usually deep red or bluish red in colour.[3] Hemorrhage may occur either spontaneously or after minor trauma. They are generally painless. Periodontally these lesions often appear to arise from the interdental gingival papilla and to spread laterally to involve adjacent teeth.[15]

Diascopy is the technique of applying pressure to a suspected vascular lesion to visualize the evacuation of coloration, a finding that supports the fact that patent blood-filled spaces constitute the lesion[5] and hemangioma may blanch on the application of pressure. Soft tissue hemangiomas may produce a cupped-out type of resorption of the bony cortex. Although these lesions do not undergo involution, they develop calcifications via phlebolith formation. These dystrophic calcifications in organized thrombi are often first seen on a radiograph. Those that occur in the parotid region are well-known to show multiple small, round radiodensities superimposed over the ramus and posterior body.[16]

Pathogenesis and origin of haemangioma remain incompletely understood. However, various theories have been proposed to elucidate the mechanism and pathogenesis of haemangioma. Aberrant and focal proliferation of endothelial cells results in haemangioma, although the cause behind this remains unclear.[17] The placental theory of haemangioma origin has been described by North et al.[18] who studied various histology and molecular markers such as GLUTI, Lewis Y Antigen, Merosin, CCR6, CD15, IDO, FC, and gamma Receptor II. Positive staining for GLUTI is considered

highly specific and diagnostic for haemangioma, and it is useful for making differential diagnosis between haemangioma and other vascular lesions clinically related to it.[17]

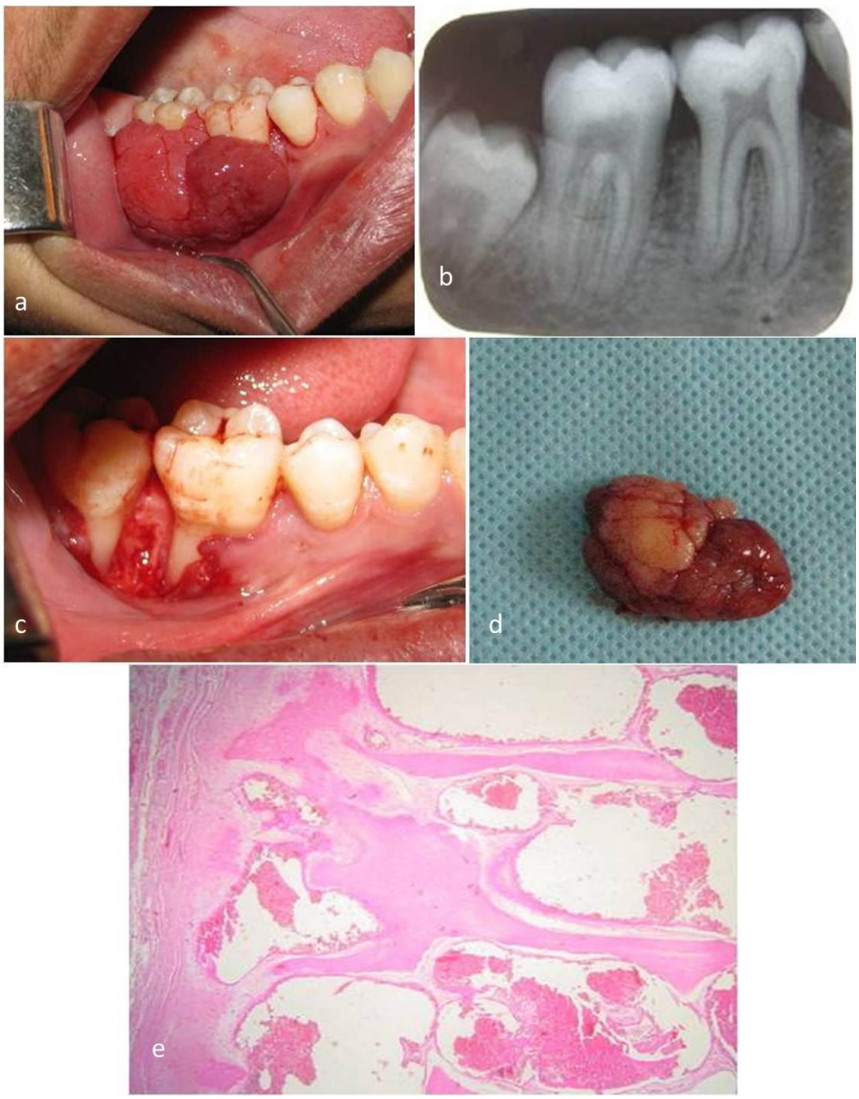

Figure 1: The Preoperative clinical view (a). Periapical radiography showing a faint radiographic shadow of the crestal bone between the first and the second right mandibular molar (b) with view immediately after the excision of the lesion showing bone loss on the buccal surface of first and second molar roots (c)**.** The excised tissue (d) with histopathological specimen consisting of hemangioma of both capillary and cavernous components (e).

More recently, somatic mutational events in gene involved in angiogenesis are related to haemangioma growth.[19] Growth factors specifically involved in angiogenesis such as VEGF, b-TGF, and IGF are often increased during the proliferation phases of haemangioma growth.[20,21] Moreover, it has been noted that during the involution phase of haemangioma, there is decrease in angiogenic molecules (VEGF, PCNA, Type IV collagenase, Lewis V antigen, CD 31), while there is increase in concentration of marker for apoptosis (T4, TUNNEL, INF, Mast cells, and TGF).[19,22,23] Thus, role of molecular signalling is now clear in haemangioma development.

Histologically hemangiomas may be classified as either capillary or cavernous; however, both types consist of proliferative vascular channels that are lined by endothelium and lack a muscular coat. Erythrocytes are in the lumen. The arrangement is often lobular, since capillaries proliferate around a feeder vessel.[16] Capillary hemangiomas may initially be extremely cellular lesions composed of endothelial cells and poorly canalized vessels. Mitoses may be present.

In early stages, these have been called juvenile hemangiomas. Mast cells, which may be a source of angiogenic factors, can be seen. As these lesions mature, the vessels are canalized, and the endothelial cells flatten to form the typical capillary hemangioma. When they undergo regression it is through interstitial fibrosis.[16]

Cavernous hemangiomas are less circumscribed than capillary hemangiomas and have dilated vascular channels with flattened endothelium. Calcifications and formation of phleboliths occur through dystrophic calcification of organizing thrombi, but regression does not occur.[16] Mixed hemangiomas consisting of both components may also occur and according to our research, this case is the first described mixed capillary-cavernous gingival hemangioma in the English literature.[4]

Hemangiomas may mimic other lesions clinically and histopathologically. The differential diagnosis of hemangiomas includes pyogenic granuloma, chronic inflammatory gingival hyperplasia, telangiectasia and even squamous cell carcinoma. The most common vascular proliferation of the oral mucosa is the pyogenic granuloma. This is a reactive lesion that develops rapidly, bleeds easily and is usually associated with inflammation and ulceration. Clinically, it is often lobulated, pedunculated and red to purple and it may be hormone sensitive.[14] Histology of pyogenic granuloma is characterized by proliferating blood vessels that are organized in lobular aggregates although superficially the lesion frequently undergoes no specific change, including edema, capillaries dilation or inflammatory granulation tissue reaction, whereas the second type consists of highly vascular proliferation that resembles granulation tissue. Histopathologically, the capillary hemangioma exhibits a progression from a densely cellular proliferation of endothelial cells in the early stages to a lobular mass of well-formed capillaries in the mature phase, often resembling the pyogenic granuloma without the inflammatory features.[14] In addition, hemangiomas may be confused with the vascular-appearing lesions of the face or oral cavity, which may also represent the Sturge-Weber syndrome. They are often located in the territory of the branches of the trigeminal nerve. Ocular and cerebral vascular lesions may be found in such cases. These lesions may be further classified into flat, telangiectatic, stellar and senile variants.[5]

Management of oral hemangiomas varies considerably depending on the age of the patient and the extent and type of the lesions. The range of treatment includes surgery,[16] laser therapy,[24] use of sclerosing agents,[25] and embolisation.[26] Most hemangiomas in infancy should be left alone, allowing for natural involution to occur.[16] Exceptions to this may include psychological reasons and lesions at eyelids affecting vision.[16] Angiography, computerized tomography and magnetic resonance imaging are useful in delineating the vascular supply and extent of the hemangiomas when their sizes are large or when jaw bones are involved. Management of such extensive lesions often requires a team approach consisting of radiologist, maxillofacial surgeon and plastic surgeon.[26] The prognosis of hemangiomas is good as it rarely recur after adequate removal.[3] The treatment carried out in the present case was surgical excision, which had been chosen based on the size of the lesion and the provisional diagnosis which include fibrous hyperplasia. However, this could have resulted in severe bleeding if the lesion was a hemangioma involving large blood vessels. If severe blood loss is anticipated, the operation must be performed in hospital setting where blood transfusion facilities are available.

Conclusion

Hemangiomas of the oral soft tissues may mimic other lesions clinically, and in some cases even histologically. Dentists should therefore be aware of these lesions when making diagnosis and attempts at excision of apparently innocent lesions may result in serious bleeding.[13]

Acknowledgement

We would like to thank all the non-teaching and teaching staff members from the oral pathology department for their extreme help.

Author Affiliations

1.Dr.Ahmed Chkoura, Specialist in Oral Surgery, 2.Dr.Wafae El wady, Professor and Head, 3.Dr.Bouchra Taleb, Professor, Department of Oral Surgery, Faculty of Dentistry, University Mohammed V Souissi, Rabat, Morocco.

References

1. Enzinger FM, Weiss SW. Soft tissue tumors. 3rd Ed. St Louis: Mosby; 1995. 581-586 p.

2. Silverman RA. Hemangiomas and vascular malformations. Pediatr Clin North Am. 1991;38:811-34.
3. Shafer WG, Hine MK, Levy BM. A Textbook of Oral Pathology. 4th Ed. Philadelphia: W.B. Saunders Company; 1983. 154-157 p.
4. Wei SHY. Pediatric Dentistry: Total Patient Care.1st Ed. Philadelphia: Lea & Febiger; 1988. 313-330 p.
5. Greenberg M, Glick M, Ship JA. Burkett's oral medicine. 11th Ed. Ontario: PMPH; 2008. 145 p.
6. Maaita JK. Oral tumors in children: a review. J Clin Pediatr Dent. 2000;24:133-5.
7. Sato M, Tanaka N, Sato T, Amagasa T. Oral and maxillofacial tumours in children: a review. Br J Oral Maxillofac Surg. 1997;35(2):92-5.
8. Tanaka N, Murata A, Yamaguchi A, Kohama G. Clinical features and management of oral and maxillofacial tumors in children. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88(1):11-5.
9. Chen YK, Lin LM, Huang HC, Lin CC, Yan YH. A retrospective study of oral and maxillofacial biopsy lesions in a pediatric population from southern Taiwan. Pediat Dent 1998;20(7):404-10.
10. Das S, Das AK. A review of pediatric oral biopsies from a surgical pathology service in a dental school. Pediatr Dent. 1993;15(3):208-11.
11. Skinner RL, Davenport WD, Weir JC, Carr RF. A survey of biopsied lesions in pediatric dental patients. Pediatr Dent 1986;8(3):163-7.
12. Wang YL, Chang HH, Chang JY, Huang GF, Guo MK. Retrospective survey of biopsied oral lesions in pediatric patients. J Formos Med Assoc 2009;108(1):862-71.
13. Cho SY, Tang MC. Haemangioma on the Dental Alveolar Ridge Report of a Case. Hong Kong Dent J. 2004;1:37-9.
14. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2nd Ed. Philadelphia: WB Saunders; 2002. 447-9 p.
15. Carranza FA. Glickman's Clinical Periodontology. 1st Ed. Philadelphia: WB Saunders Co; 1990, 335-51 p.
16. Marx RE, Stern D. Oral and maxillofacial pathology: a rationale for diagnosis and treatment, 1st Ed. Illinois: Quintessence Publishing; 2003. 442 p.
17. Buckimler LM, Richter GT, Suen JY. Diagnosis and management of hemangiomas and vascular malformation of the head and neck. Oral Dis. 2010;16(5):405-18.
18. North PE, Waner M, Brodsky MC. Are infantile hemangiomas of placental origin? Ophthalmol. 2002;109(4):633–4.
19. Gombos F, Lanza A, Gombos F. A case of multiple oral vascular tumors: the diagnostic challenge on haemangioma still remain open. Judicial Studies Institute Journal 2008;2(1):67-75.
20. Chang J, Most D, Bresnick S, et al. Proliferative hemangiomas: analysis of cytokine gene expression and angiogenesis. Plast Reconstr Surg. 1999;103(1):1-10.
21. Kleinman ME, Greives MR, Churgin SS, et al. Hypoxia induced mediators of stem/progenitor cell trafficking are increased in children with hemangioma. Arterioscler Thromb Vasc Biol. 2007 Dec;27(12):2664-70.
22. Frischer JS, Huang J, Serur A, Kadenhe A, Yamashiro DJ, Kandel JJ. Biomolecular Markers and Involution of Hemangiomas. J Pediatr Surg. 2004;39(3):400-4.
23. Sun ZJ, Zhao YF, Zhao JH. Mast cells in hemangioma: a double-edged sword. Med Hypotheses. 2007;68(4):805-7.
24. Barak S, Katz J, Kaplan I. The CO2 laser in surgery of vascular tumors of the oral cavity in children. ASDC J Dent Child. 1991;58(4):293-6.
25. Chin DC. Treatment of maxillary hemangioma with sclerosing agent. Oral Surg Oral Med Oral Pathol. 1983;55:247-9.
26. Greene LA, Freedman PD, Friedman JM, et al. Capillary hemangioma of the maxilla. A report of two cases in which angiography and embolization were used. Oral Surg Oral Med Oral Pathol 1990;70(3):268-73.

Corresponding Author

Dr.Ahmed Chkoura,
Specialist in Oral Surgery,
Department of Oral Surgery,
Faculty of Dentistry,
University Mohammed V Souissi,
Rabat, Morocco.
Email:

Source of Support: Nil, Conflict of Interest: None Declared.