Intraoral Capillary Hemangioma in Mandibular Anterior Region – A Rare Entity: A Case Report

Sara Siddiqua*, Abdul Khader, Mohsin Ali, Arjun Goud, Mandula Nagesh, Meraj Sultana, G. G. Sheela Prakash
Department of Oral and Maxillofacial Surgery, Sri Balaji Dental College and Hospital, Himayatnagar, Telangana, India.

Abstract

Background: Hemangiomas are common benign vascular tumors of the head and neck region. They have a female predilection. On Microscopic appearance, they are classified into capillary, cavernous, mixed, and sclerosing variety, Incidence of intraoral capillary hemangioma (CH) is infrequent, and its topographical presentation on the oral mucosa marks extreme rarity.

Keywords: Oral hemangioma (OH); Capillary hemangioma (CH); Vascular endothelial, Growth factor (VEGF); Glucose transporter 1 (GLUT1), International Society for the Study of Vascular Anomalies (ISSVA).

Introduction

Hemangioma is a relatively common benign proliferation of blood vessels that primarily develops during childhood, 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 and uncommonly encountered by clinicians.[1,2] They might be cutaneous, affecting the skin, lips, and deeper tissues; mucosal, affecting the oral cavity lining; intramuscular, affecting the masticator and perioral muscles; or intraosseous, affecting the mandible and or maxilla.[3]

Two main forms of hemangioma are recognized as capillary and cavernous. A very few cases of intraoral CHs have been reported in the literature.[4] In 1973 Sznajder *et al*. were the first to describe it in the literature, under the term "Hemorrhagic hemangioma" The capillary form presents as a flat area consisting of numerous small capillaries. In microscopic examination, Capillary hemangioma (CH) is a kind of intraoral neoplasm that consists of multiple tiny capillaries lined by a single layer of endothelial cells and supported in a connective tissue stroma of different densities.[5] CHs account for 0.5–1.0% of all intraoral neoplasms.[6]

Case Presentation

A 60-year-old female was referred to our department of oral and maxillofacial surgery for evaluation and treatment of the gingival overgrowth and bleeding that she suffered during meals since 1 month accompanied by an elevated reddish discolored gingiva, a few days later, she discovered a dark red swelling on her lower anterior alveolar mucosa. This swelling had been increasing gradually since that time. She did not provide any information about her dental history. The patient's medical history was non-contributory and she was not on any medications.

A comprehensive intraoral examination revealed a localized gingival mass occupying the missing mandibular right central incisor and left incisor (41, 31,32,33) (Figure 1). It was firm, pedunculated with a distinct stalk arising from the interdental papillary region. The mass was bright red, erythematous, and single lobulated with well-defined margins and measured about unilateral growth of 2.5 cm × 2.5 cm (Figure 1). It was firm and rubbery in texture with no surface ulceration or secondary infection noted. Periodontal examination revealed no clinical attachment loss. Panoramic examination confirmed no alveolar bone loss (Figure 2), and a provisional diagnosis of pyogenic granuloma was made on the basis of history and clinical features.

A excisional biopsy was planned, the lesion was excised in total, that produced profuse hemorrhage which by digital compression was uncontrolled, electric cauterization was

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Address for correspondence: Dr. Sara Siddiqua, Department of Oral and Maxillofacial Surgery, Sri Balaji Dental College and Hospital, Himayatnagar - 500 075, Telangana, India.
Email: z3iy4321@gmail.com

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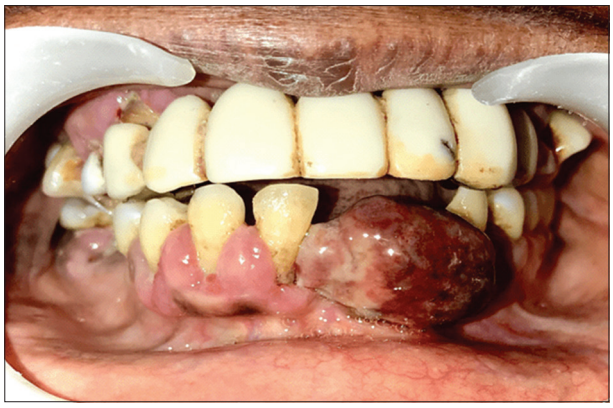

Figure 1: Pre-operative. A localized gingival mass occupying the missing mandibular right central incisor and left incisor (41, 31, 32, and 33)

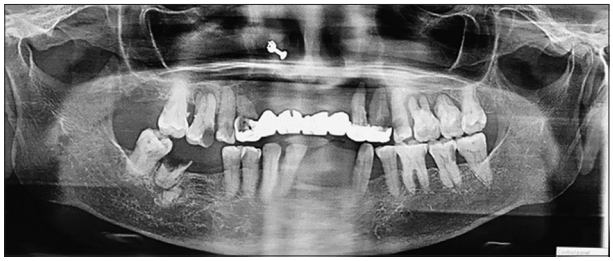

Figure 2: Pre-operative. Oral Panoramic graph shows shadow of the growth in mandibular anterior region with no alveolar bone loss

done to achieve hemostasis (Figure 3). The biopsied tissue was rinsed in 10% formalin and sent for histopathologic examination. Histopathologic examination of the excised tissue revealed nonkeratinized stratified squamous epithelium overlying on an encapsulated tumor composed of many thin-walled capillary channels. A single layer of endothelial cells coated the capillaries. Endothelial cell growth was seen in some locations. Throughout the stroma, there were a few plasma cells and lymphocytes (Figure 4).

Discussion

Hemangiomas are tumors identified by rapid endothelial cell proliferation in early infancy, followed by involution over time; vascular malformations result from anomalous development of vascular plexuses.[2] Hemangiomas are usually classified into capillary, cavernous, and mixed hemangioma, difference between capillary and cavernous hemangioma is that CH has numerous proliferating small thin-walled blood filled vessels where as in cavernous hemangioma, there are deep irregular and dermal tangles of large thin-walled vessels or sinusoids, CH are surrounded by discontinuous layer of pericytes and reticular fibers, cavernous hemangioma is surrounded by discontinuous layer of endothelial cells, CH has single layer of flattened or plump endothelial cells and cavernous hemangioma is separated by scanty connective tissue.[7]

CH consists of proliferative vascular channels that are lined by endothelium and lack a muscular coat. The arrangement is usually lobular, since capillaries proliferate around a feeder

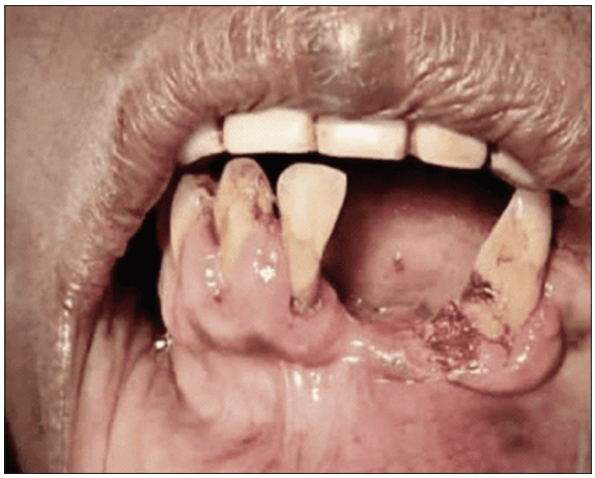

Figure 3: Intraoperative. A excisional biopsy was planned, the lesion was excised in total, producing perfused uncontrolled hemorrhage. Electric cauterization was done in order to achieve hemostasis

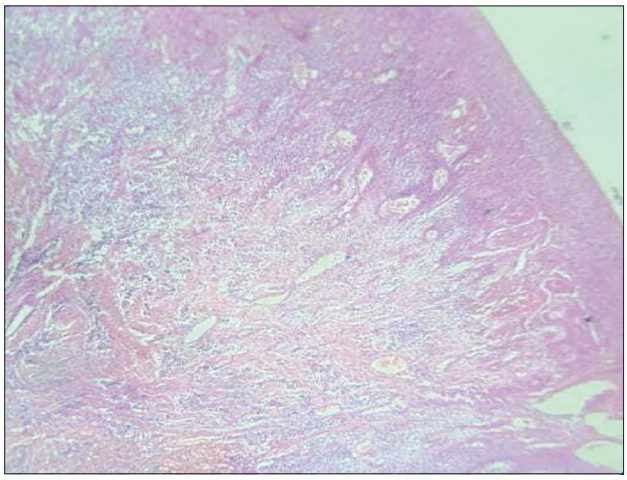

Figure 4: Histopathological image under 4× magnification shows Parakeratinized stratified squamous epithelium, with fibrous connective tissue stroma consisting of varied sized numerous blood filled capillaries can be seen

vessel. CH composed of endothelial cells that poorly canalized vessels.[3]

CH s may mimic other lesions clinically, radiographically, and histopathologically. The differential diagnosis of hemangiomas includes pyogenic granuloma, chronic inflammatory gingival hyperplasia (epulis), epulis granulomatosa, varicocele, telangiectasia, and even with squamous cell carcinoma.[2] The most common vascular proliferation of the oral mucosa is the pyogenic granuloma, this is a kind of reactive lesion that grows quickly, bleeds readily, and is often accompanied by inflammation and ulceration.[4] Clinically, it is often lobulated, pedunculated, and red to purple and it may be hormone sensitive.[5]

CHs are composed of small thin-walled capillary size that are lined by a single layer of flattened or plump endothelial cells and surrounded by a discontinuous layer of pericytes and

reticular fibers.[8] CH consists of an array of capillary-sized blood vessels with lobular architecture.[4] Such similar findings was noted in the present case report (Figure 4).

According to the literature, CH is a frequent soft-tissue tumor of the head and neck, but it is a relatively unusual and uncommon pathologic entity in the oral cavity.[1] CH with its major site on the gingiva appears to be quite uncommon. The most frequent site of occurrence of intraoral hemangiomas are lips, tongue, buccal mucosa, and palate. It is very rare to find hemangioma on attached gingiva.[9,6]

The ISSVA modified it in their continuing workshops, differentiating vascular tumors from vascular malformations based on their clinical appearance, radiological features, pathological features, and biological behavior, vascular tumors are classified as infantile hemangiomas, which can be classified as focal, segmental, and indeterminate, the other variants are congenital hemangiomas, hemangiomas which are rapidly involuting, congenital, hemangioma, non-involuting, congenital, hemangioma, other than that Tufted angioma, pyogenic granuloma, dermatologic acquired, vascular tumors, Kaposiform, hemangiomas which are endothelial in origin; hemangioendothelioma, spindle cell, comes under vascular tumors, vascular malformation can be classified as slow (low) flow malformation which are classified as capillary malformations, port-wine stain, telangiectasia, and angiokeratoma, venous malformations (VM) are classified as common sporadic VM, Bean syndrome, Familial cutaneous and mucosal venous malformations capillary malformations, Glomuvenous malformation, and Maffucci syndrome, lymphatic malformation is classified as lymphedema, lymphangioma circumscriptum, lymphangioma cavernosum, and lymphangiona cysticum, fast (high) flow malformation are arterial malformation, arteriovenous fistula/Arteriovenous malformation, and complex combined vascular malformation.[10]

Different treatment procedures used in treating hemangiomas include microembolization, radiation, cryotherapy, sclerosing agents, surgical excision, and recently Erbium lasers have been used.[7]

In the case presented here, when the surgical excision was done, uncontrolled bleeding was encountered, for which electric cautery used for cauterization, hemostasis was achieved, the patient was kept under observation, and antibiotics were prescribed, the patient was recalled at regular intervals and no sign of recurrence was reported until 6 months of follow-up.

Conclusion

The present case is important because of its rarity, its location, its occurrence in middle aged individual, this type of lesion is not commonly encountered in regular practice and clinician does not expect this type of oral lesion;, however, while treating such lesions, various complication may occur, risk of bleeding is always a main complication while dealing these kind of lesions, so clinician should be aware and acquire all the data, evaluate the patient thoroughly, the aim of this article is to highlight such lesion.

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