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Case Report

Atypical Lobular Capillary Hemangioma as an Unusual Presentation in Mandible: Case Report and Literature Review

Nicolás Solano, Ligia Pérez, Valentina Gutiérrez, Paulina Gutiérrez, Betsabe Sarcos*, Ana Villarroel and Ariamay Castrillo

Oral Surgery Post-Graduated Program, School of Dentistry. Universidad del Zulia, Venezuela

***Corresponding Author:** Betsabe Sarcos, Oral Surgery Post-Graduated Program, School of Dentistry. Universidad del Zulia, Venezuela.

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Abstract

Lobular capillary hemangioma (LCH), commonly described in the literature as "Pyogenic granuloma" is a reactive tumor-like lesion commonly affecting the oral cavity closely related to trauma, chronic irritation and hormonal changes. Clinically, the lesion appears as a smooth, lobulated, exophytic mass, exhibiting pink to reddish-purple in color and usually easy bleeding. The size of lesion varies from millimeters to centimeters, rarely exceeding 2 - 2.5 cm. We report a clinical case of lobular capillary hemangioma of the mandible in a male patient with unusual clinical presentation and imaging features.

Keywords: Oral Pyogenic Granulomas; Lobular Capillary Hemangioma; Reactive Lesion; Vascular Lesion

Introduction

Lobular capillary hemangioma (LCH), commonly described in the literature as "Pyogenic granuloma" represents benign vascular tumors that affect skin and mucous membranes. Pyogenic granuloma of the oral cavity most frequently affects the gingiva, but these lesions can be seen on the lips, tongue, buccal mucosa and palate presenting as a nodular growth that can be slow or fast in nature [1,2]. These lesions are characterized by a proliferation of capillaries arranged in lobes and separated by a lax connective tissue stroma, often infiltrated by inflammatory cells [3]. The most common triggers of pyogenic granuloma are trauma, chronic irritation, drugs, hormonal imbalances, faulty restorations, which leads to excessive proliferation of vascular connective tissue [4]. The objective of this manuscript was to report a clinical case of lobular capillary hemangioma of the mandible with unusual clinical presentation and imaging features and to review the literature regarding this pathology.

Case Report

A 54-year-old male patient attended the Oral Surgery and Maxillofacial Unit of Hospital Coromoto - Venezuela, showing increased volume in the right side of the face, with approximately 6 months of evolution, asymptomatic, without treatment. The patient has a history of arterial hypertension treated with Carvedilol 25 mg and Losartan 50 mg. Intraorally, an exophytic, pedunculated lesion was observed, lobulated, firm, erythematous, bleeding with ulcerated areas, with a size of 4 x 5 cm approximately located at the mandibular alveolar ridge level on the right side, it caused grade II dental mobility of the adjacent second molar, similarly, expansion of the corticals was evidenced from mandibular symphysis extending to its body and ipsilateral angle. Complementary studies were performed and a panoramic radiography and cone-beam computed tomography showed compatible images of an osteolytic process, which involved symphysis, body and mandibular angle on the right side of the face, with hyperdense images suggestive of locoregional periosteal response in the mandibular body. Following the Unit protocols established an incisional biopsy of the exophytic soft tissue lesion after aspiration was performed. The sample was sent to histopathological study, where a stroma of fibrous connective tissue was evidenced, occupied by spindle cells, some with slightly pleomorphic and hyperchromatic nuclei, organized in fascicles, a

storiform pattern in some areas, diagnosing spindle-shaped cell neoplasm. Immunohistochemical study showed proliferation of congestive blood vessels surrounded by edematous stroma with inflammatory cells and marked proliferation of reactive stromal myofibroblasts. The blood vessel wall was immunoreactive to Vimentin, CD34, and Smooth muscle actin (SMA), confirming the Lobular capillary hemangioma diagnosis. Subsequently, it was decided to take the patient to surgery under general anesthesia, for excisional biopsy, where the exophytic lesion located in the right mandibular alveolar ridge was resected and enucleation and curettage of the central mandibular lesion was performed. Postoperative controls were performed during 6 months without signs of recurrence.

Figure 1: Intraoral photograph showing nodular, lobulated, exophytic swelling, with purplish-red color and whitish areas of approximately 3 × 4 cm, present in alveolar ridge of the mandible on the right side.

Figure 2: Panoramic radiograph showing a radiolucent, multilocular image that covers symphysis, body and mandibular angle on the right side.

Figure 3: Cone beam computed tomography axial section showing osteolytic process that involves symphysis and mandibular body on the right side with the presence of periosteal reaction.

Figure 4: Histopathological image showing a fibrous connective tissue stroma occupied by spindle cells organized in fascicles.

Figure 5: Immunohistochemical study where it is reported that the vessel wall is reactive to: a) smooth muscle actin 400x and b) CD 34. 400x.

Discussion and Conclusion

Pyogenic granuloma is a common tumor of the oral cavity considered non neoplastic in origin. In 1904 Hartzell added the term "Pyogenic granuloma" or "granuloma pyogenicum"; however, this term is considered inappropriate due to the fact that the pyogenic granuloma doesn't produce pus and doesn't represent a true granuloma [4,5]. According to the literature, oral pyogenic granulomas show a notable predilection for the gingiva, representing 75% of all cases. The lips, tongue and oral mucosa are the next most common sites [6]. When it occurs in the intraoral mucosa during pregnancy, especially in the gums, it is called granuloma gravidarum, granuloma of pregnancy or epulis gravidarum, generally in the second or third trimester of gestation [7]. Although pyogenic granuloma can develop at any age, it is more common in children and young adults. Most studies also show a female predilection, possibly due to the vascular effects of female hormones [6]. In the reported case, the lesion diagnosed as pyogenic granuloma occurred in a male patient in the fifth decade of life, being a rare age of appearance of that lesion, therefore, it wasn't among the first differential diagnoses. According to its vascularity, pyogenic granuloma can be classified as lobular capillary hemangioma type and non-lobular capillary hemangioma type, both different histologically and clinically [8,13]. Among the common characteristics of LCH, solitary, compressible and painless masses are reported, that can vary from pink to red and purple, depending on evolution time. This injury can cause ulceration and constant bleeding. The etiology is unknown, however, trauma such as bites, dental fractures, por restorations, chronic local irritation, hormonal factors such as pregnancy or oral contraceptives, poor oral hygiene and medications can predispose the development of these injuries [5,8]. The 54-year-old male patient presented an exophytic lesion, firm, bleeding with an intense red to purplish color, showing some ulcerated areas, with an approximate size of 3 x 5 cm located at the alveolar ridge of the mandible level on the right side, causing the mobility of dental organ number 47, being these characteristics commonly reported in this type of pathology.

Radiographically this type of lesion does not present bone involvement but in some cases may show slight superficial erosion at the bone level and the extensive loss of alveolar bone and tooth mobility is observed in 3% of patients [9]. In accordance with what is reported in the literature, this case showed imaging characteristics of a central, multilocular lesion, with expansion and fenestration of the bone cortices of the mandible, these being characteristics reported only in 3% of cases. Its presentation simulated an aggressive lesion with a possible malignant nature for which pathologies such as ameloblastoma and osteosarcoma were considered as differential diagnoses. Histopathologically, the pyogenic granuloma shows a lush granulation tissue that is covered by atrophic and hyperplastic epithelium that may ulcerate sometimes and reveals fibrinous exudates, numerous endothelium-lined vascular spaces and proliferation of fibroblasts and endothelial cells are evident [10]. The immunohistochemical study can confirm the diagnosis of pyogenic granuloma when several specific markers such as CD-34, CD-68 are present [11]. In this case, the microscopic findings showed fibrous connective tissue stroma, occupied by spindle cells, some with slightly pleomorphic and hyperchromatic nucleus, organized in fascicles, in a storiform pattern in some areas suggesting spindle cell neoplasia, so an immunohistochemistry study was performed to specify the diagnosis observing proliferation of congestive blood vessels surrounded by edematous stroma rich in inflammatory cells and marked proliferation of myofibroblast reactive stromals. The blood vessel wall was immunoreactive to Vimentin, CD34 and Smooth muscle actin (SMA), confirming the lobar capillary hemangioma diagnosis. Since the pyogenic granuloma is a benign lesion, complete surgical excision is the treatment of choice. The recurrence rate is around 16% of excised lesions, to prevent the possibility of recurrence due to chronic irritation, the lesion should be excised down to the periosteum along with scraping and root planing of adjacent teeth [12,13]. In the presented case, complete excision of the exophytic lesion was performed, extraction of the dental organ 47, plus enucleation and curettage of the central lesion in the mandible that extended from the mandibular symphysis up to the mandibular angle on the right side. The LCH type is reported to have a lobular arrangement of capillaries and

proliferating blood vessels. This proliferation of blood vessels occurred in our case where the blood vessels were arranged in the path of the injury at the time of excisional biopsy, correlating in the same way with the histopathological results of the biopsy. Many other reports approved in the literature of this entity make clear the characterization of it, however the manifestation of this lesion compromising unusual anatomical areas such as the one reported in this case is of great interest, because due to its characteristics it can simulate other clinical entities of aggressive or even malignant etiology, for this reason its identification and proper diagnosis are very important in order to indicate the right treatment for this pathology and avoid unnecessary aggressive therapy.

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