

Crocker & Hartzell's Disease of the Tongue: Two Case Reports with Review of Literature

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Abstract

Crocker and Hartzell's disease is an inflammatory hyperplasia of the skin and oral mucosa. This lesion is not associated with pus and histologically it resembles an angiomatic lesion rather than a granulomatous lesion. It is considered as a non-neoplastic tumor, and it presents itself in the oral cavity in various clinical and histological forms. The most common site of occurrence in the oral cavity is the gingiva. In this article, we are presenting two cases of Crocker and Hartzell's disease occurring on the tongue at two different locations. A 52-year-old female patient with a lesion on the dorsum of the tongue, and a 53-year-old male patient with the lesion on the lateral border of the tongue.

Key words: Disease, Gingiva, Non-neoplastic, Pyogenic granuloma, Tongue

INTRODUCTION

Crocker and Hartzell's disease is a synonym for pyogenic granuloma which is otherwise also known as "Capillary hemangioma"¹ or "granuloma pyogenicum"² due to the presence of numerous blood capillaries. It is an inflammatory hyperplasia of the skin and mucous membrane. First case of pyogenic granuloma was reported in 1844 by Hullihen and the term was coined by Hartzell in 1904.³ It occurs due to chronic low-grade trauma, physical trauma, hormonal factors, bacteria, viruses, certain drugs, calculus, and poor oral hygiene. The gingiva is the most common site of occurrence, accounting for 75% of all cases.⁴ It is a non-neoplastic tumor of the oral cavity and has got many clinical and histological forms. The term itself is considered as a misnomer as it is not associated with pus clinically and histologically it is an angiomatic lesion rather than a granulomatous lesion.⁵

In this article, we have presented two case reports of pyogenic granuloma occurring on the tongue. One case

of the 52-year-old female patient who came with an asymptomatic lobulated and pedunculated swelling on the dorsum of the tongue; and another case of the 53-year-old male patient who came with an asymptomatic non-lobulated sessile swelling on the lateral aspect of the tongue. The etiological factor was different in both the cases as the female patient had a poor oral hygiene and the male patient had sharp lingual cusps of the teeth that were lying in close approximation with the tongue.

We have also reviewed the literature and discussed the present cases with reference to the same.

CASE REPORT

The 52-year-old female patient came to the Department of Oral Medicine and Radiology with a chief complaint of a swelling on the dorsum of the tongue which she has been noticing for the past 6 months. She also gave a history of bleeding from the swelling while brushing and while having food. She also reported that the swelling was initially small in size which later increased to the present size.

On intraoral examination (Figure 1) there was the presence of an exophytic, pink, lobulated and pedunculated lesion measuring about 1 cm in diameter with a pseudomembranous surface with areas of erythema. The oral hygiene status was poor with a few missing teeth. The lesion was soft

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in consistency and non-tender with minimal bleeding. The provisional diagnosis was pyogenic granuloma with a differential diagnosis of irritational fibroma.

In another case, the 53-year-old male patient came to the Department with a chief complaint of a swelling in the right side of the tongue since the last 3 months. The patient experienced mild pain and bleeding while having food.

On intraoral examination (Figure 2) there was the presence of a sessile swelling of approximate size $1\text{ cm} \times 0.5\text{ cm}$ on the right lateral border of the tongue almost 4-5 cm behind the tip of the tongue. The surface seemed to be mildly erythematous. On palpation, the lesion was firm in consistency and mild tenderness was present. Sharp lingual cusps in relation to 45, 46 were observed. In this case also we considered pyogenic granuloma as a provisional diagnosis.

Both the patients were diabetic and were being prepared for biopsy with written consent after routine blood

investigations and blood sugar examinations. Excisional biopsy was done under local anesthesia, and the specimens were sent to the department of oral pathology for histological examination.

The histopathological diagnosis was given as pyogenic granuloma in both the cases.

DISCUSSION

The exact etiology of this lesion is unknown though it was originally believed to be a botryomycosis infection.³ Regezi *et al.*⁴ suggested that pyogenic granuloma can be caused by any stimulant or an injury such as calculus or foreign material and Ainamo² suggested that routine tooth brushing causes repeated trauma to the gingiva resulting in irritation and exuberant proliferation of these connective tissue lesions. Trauma to deciduous dentition, abnormal tooth development, occlusal interferences, and immunosuppressive drugs such as cyclosporine and improper placement of healing cap for implants are some of the other precipitating factors for pyogenic granulomas.² Kerr *et al.* has reported that staphylococci and botryomycosis cause localization of infection in blood vessel walls which contribute to the formation of the lesion.³

In our patients, the lesion may have occurred in the lady due to the poor oral hygiene and in the male due to the chronic trauma of a sharp cusp on the lateral border of the tongue.

Oral pyogenic granulomas occur in all age groups, from young to the old, but are the most frequent encountered in females in their second decade due to the increased levels of circulating hormones estrogen and progesterone.² In our case both patients were in their fifth decade of life. Hosseini *et al.*,² observed an increase in gingival enlargements during pregnancy and atrophied cases in menopause. Yuan *et al.*,² reported that the morphogenetic factors were higher in pyogenic granuloma rather than normal gingiva supporting the mechanism of angiogenesis in oral pyogenic granulomas in pregnant females. However, the effects of female hormones on oral pyogenic granulomas were questioned by Bhaskar and Jacoway⁵ since they found lesions both in males and females and concluded that there is no specific sex predilection. Even in our case both the sexes have been affected.

Bhaskar and Jacoway⁵ also demonstrated the presence of Gram-positive and Gram-negative bacilli in the ulcerated form of pyogenic granuloma suggesting that these organisms are contaminants from the oral cavity. This probably justified the inclusion of the term "pyogenic" in the term pyogenic granuloma which otherwise shows prominent capillary growth within a granulomatous mass

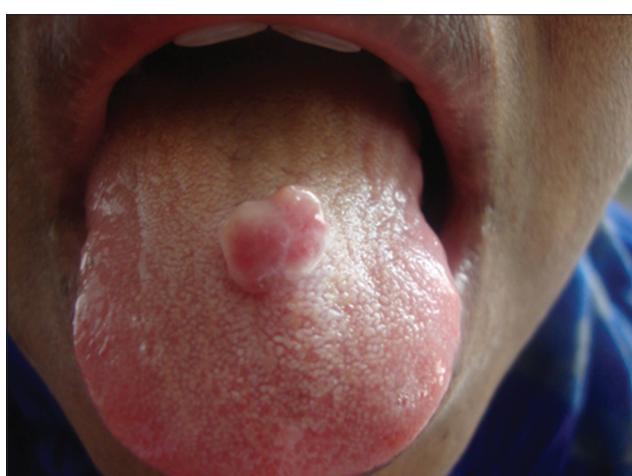


Figure 1: A lobulated and pedunculated swelling on the dorsum of the tongue of the 52-year-old female patient

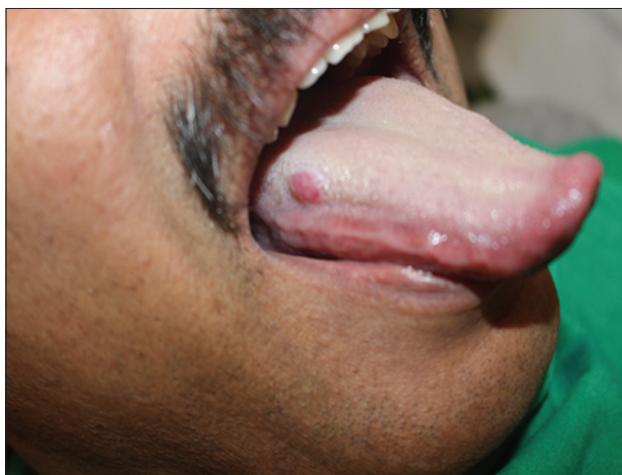


Figure 2: A smooth surfaced sessile lesion on the lateral border of the tongue in the 53-year-old male patient

rather than the presence of pyogenic organisms and pus, so the term itself is a misnomer suggesting that it is not a granuloma in the real sense.⁴

The pyogenic granuloma of the oral cavity appears as an elevated, smooth or exophytic, sessile, or pedunculated growth covered with red hemorrhagic, and compressible erythematous papules, which appears lobulated and warty showing ulcerations and at times covered by yellow fibrinous membrane.⁴ There are two kinds of pyogenic granuloma namely lobular capillary hemangioma and non-lobular capillary hemangioma type, which can be differentiated by their histological features.¹ In case 1, the lesion was pedunculated and exophytic in nature. In case 2, the lesion was sessile and erythematous in nature.

The color varies from pink, red, or reddish purple depending on the vascularity of the growth.² Young pyogenic granulomas are highly vascular as they are composed predominantly of hyperplastic granulation tissue with capillaries.¹ Thus, even minor trauma may cause considerable bleeding in such lesions. Whereas older lesions become more collagenized and pink.⁷ In our case, both the lesions were erythematous due to trauma from the adjacent teeth.

The most common site of occurrence is the gingiva. Besides the gingiva it can also be noticed on the lips, tongue, or buccal mucosa, affecting the maxilla more than the mandible, the anterior region more than the posterior with the buccal surfaces being affected more than the lingual surfaces.⁸ In our case, both the lesions have occurred on the tongue stressing on the fact that it has occurred at two different sites.

The size of the lesion varies from a few millimeters to several centimeters in diameter. It is usually a slow asymptomatic growth,⁶ but at times may grow rapidly.² In our case, both lesions were approximately 1 cm in size.

Over the years various authors have suggested synonyms for the lesion such as granuloma gravidarum, pregnancy tumor, Crocker and Hartzell's disease, vascular epulis, benign vascular tumor, hemangiomatosis granuloma, epulis teleangiectaticum granulomatosa, and lobular capillary hemangioma.⁷ Kelley and Bernard regarded pyogenic granuloma as a "benign, acquired, vascular, and neoplasm."⁹

Although pyogenic granuloma is diagnosed clinically, radiographic and histopathological investigations aid in confirming the diagnosis and treatment. Radiographs are advised to rule out bony destructions suggestive of malignancy or to identify a foreign body.¹⁰

Histopathologically pyogenic granuloma is characterized by stratified squamous surface epithelium with underlying

connective tissue showing the proliferation of endothelial cells in a lobular pattern with numerous capillaries.⁸ In both our cases also similar histopathological findings were found (Figures 3 and 4).

The differential diagnosis for pyogenic granuloma included peripheral giant cell granuloma, peripheral ossifying fibroma, metastatic cancer,⁶ hemangioma, pregnancy tumor, conventional granulation tissue hyperplasia, Kaposi's sarcoma, bacillary angiomatosis, and non-Hodgkin's lymphoma.² The peripheral giant cell granuloma can be histologically identified due to the presence of multinucleated giant cells.⁶ Ossifying fibroma or peripheral odontogenic fibroma occurs exclusively on the gingiva; however, it has a minimal vascular component compared to pyogenic granuloma.^{1,4} Due to the proliferating blood vessels differential diagnosis of pyogenic granuloma from a hemangioma is made histologically in which hemangioma shows endothelial cell proliferation without acute inflammatory cell infiltrate,² which is a common finding in pyogenic granuloma. The

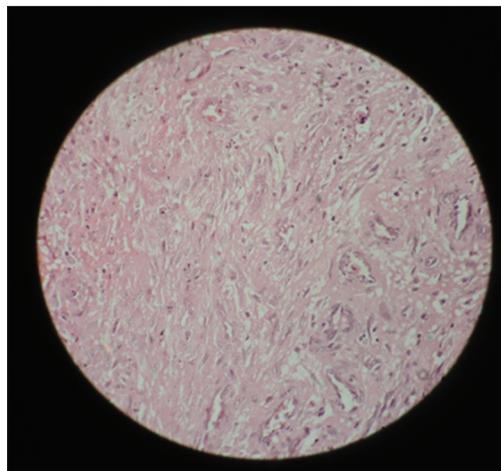


Figure 3: Histopathology picture of the 52-year-old female patient

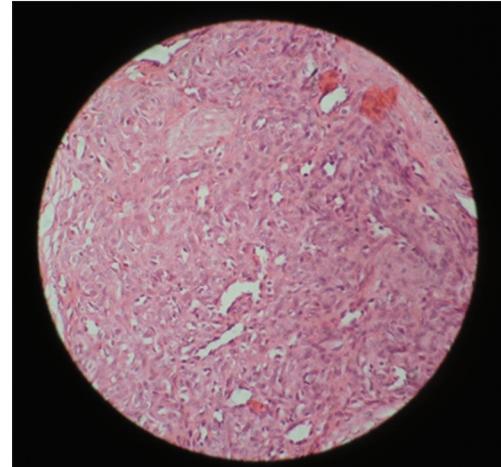


Figure 4: Histopathology picture of the 53-year-old male patient

diagnosis of pregnancy tumor is based on history and the influence of the female sex hormones.² Conventional hyperplastic gingival inflammation resembles pyogenic granuloma in histopathologic sections, and it is impossible for the pathologist to reach a diagnosis and in such cases.² The pyogenic granuloma is distinguished from Kaposi's sarcoma in acquired immunodeficiency syndrome due to the proliferation of dysplastic spindle cells, vascular clefts, extravasated erythrocytes, and intracellular hyaline bodies² none of which are seen in pyogenic granuloma.

The treatment for pyogenic granuloma includes excisional biopsy and for larger lesions incisional biopsy is recommended.⁴ Conservative surgical excision of the lesion with removal of irritant such as plaque, calculus, and foreign materials is recommended for small painless non-bleeding lesions.

Various other treatment modalities such as use of neodymium-doped yttrium aluminium garnet laser, carbon dioxide laser, flash lamp pulse dye laser, cryosurgery, electrodesiccation, and sodium tetradecyl sulfate sclerotherapy⁵ and use of intralesional steroids² have been used by various clinicians.

Even our two cases, an excisional biopsy was being done. When the patient was being reviewed after 1 week, the biopsy site had completely healed, and we continued the follow-up for 1 month and 6 months (Figure 5).

Taira *et al.*,⁴ have shown a recurrence rate of 16% of cases. Incomplete excision, failure to remove etiologic factors or repeated trauma contributes to recurrence of these lesions.¹ Vilmann *et al.*,² emphasized the need of follow-up for the cases.

CONCLUSION

Crocker and Hartzell's disease or pyogenic granuloma is a common lesion of the skin and oral cavity, especially the gingiva. The cases of pyogenic granuloma occurring in an extra gingival region such as on the tongue in a male and female patient in the fifth decade of life gives an insight into its myriad etiological factors, clinical features and appearances, histological presentations, treatment

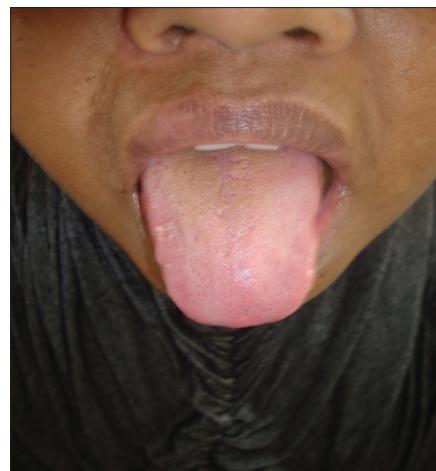


Figure 5: Complete recovery of the lesion after a follow-up of 1 month

modalities, and recurrence rates and describes how the diagnosis and treatment of such a case was completed and followed up for a period of 6 months. The article also highlights the fact that though the term pyogenic granuloma is frequently used it is not associated with pus, and histologically it resembles angiomatic lesion rather than granulomatous lesion indicating that the term "pyogenic granuloma" is a misnomer.

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