CASE REPORT

A Non-Classical Presentation of Oral Pyogenic Granuloma with Actinomycosis

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Abstract

Introduction: Pyogenic granuloma is a rather confusing reactive lesion of the oral mucosa, considering it is neither a bacterial infection nor does it produce any pus. In fact, even histologically, there is no formation of granulomas to substantiate the name. Intraoral actinomycosis is another interesting lesion due to its unique ability to masquerade itself as a swelling, abscess, or even a neoplasm. The occurrence of the two lesions separately is common in the oral cavity, however, their co-existence is extremely rare.

Case Presentation: A 65-year-old female patient was referred from a private dental clinic with the chief complaint of a growth on her left posterior region of mouth, at the occlusal level of 15, 18 tooth regions for past 1 month.

Conclusion: Clinicians should be aware about the occurrence of such rare combination of intraoral lesions, in order to make an accurate diagnosis and in turn, tailor a correct treatment protocol for these patients.

Keywords: Pyogenic Granuloma, Actinomycosis, Bacterial infection

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Introduction

Pyogenic granuloma can be described as a reactive vascular lesion of inflammatory nature that arises as a response to various stimuli such as chronic local irritation, traumatic injury or hormonal changes. It commonly presents as a soft red, pinkish or purplish solitary growth of the oral cavity and skin. The colour of the lesion is varied and depends on its vascularity and clinical course namely, "early", "established" or "healing".[1] Coined by Hartzell in 1904, the term pyogenic granuloma is rather confusing, considering it is neither a bacterial infection nor does it produce any pus.[2] In fact, even histologically, there is no formation of granulomas to substantiate the name.

Actinomycosis, on the other hand, is a chronic granulomatous suppurating bacterial infection caused by actinomyces species, which are otherwise normal commensals of the human oral cavity. It is a slow progressive infection which manifests as multiple abscesses and sinus tract infection.[3] Isolated actinomycosis, however, are uncommon and are usually accompanied by cervicofacial form of the disease.[4]

Though pyogenic granuloma is a common lesion occurring in the oral tissues, it's association with Actinomycosis is rare. The present case is one such and is therefore presented for its rarity.

Case Presentation

A 65-year-old female patient was referred from a private dental clinic with the chief complaint of a growth on her left posterior region of mouth, at the occlusal level of 15, 18 tooth regions (Universal system) for past 1 month. The medical and personal history of the patient was non-contributory. There was no gross facial asymmetry noticed on extra oral examination. Intraoral examination revealed a solitary, pedunculated growth, measuring approximately 1x1 cm^2, in relation to the left buccal mucosa at the occlusal level of 15, 18 tooth regions. The growth was soft in consistency with smooth surface and its colour was similar to the adjacent normal mucosa. There was no evidence of any discharge from the growth or any associated symptoms. No regional lymphadenopathy was present. The oral hygiene status of the patient was poor. A provisional diagnosis of fibroma/

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fibroepithelial polyp was made and the lesion was excised. The excised specimen was sent for histopathological examination.

Grossing showed greyish white soft tissue specimen, soft in consistency with a smooth surface. On histopathological examination, non-keratinised stratified squamous epithelium with the underlying cellular connective tissue stroma containing abundant endothelium lined blood capillaries, proliferating plump fibroblasts and budding endothelial cells were seen. Numerous meshwork of thin basophilic filamentous structures was noticed encapsulating the lesion in a palisading pattern with peripheral eosinophilic club shaped radiating ends, resembling the so called "ray fungus" (Figure 1, 2&3). Presence of fibrinopurulent area with a sea of neutrophils was seen adjacent to these filamentous structures. (Figure 4) Moderate infiltration of mixed inflammatory cells such as polymorphonuclear leukocytes, lymphocytes and plasma cells were also evident (Figure 5).

Correlating the clinical and histopathological findings, a definitive diagnosis of oral pyogenic granuloma with actinomycosis was made.

Discussion

Oral pyogenic granuloma is a common soft tissue growth of the oral cavity that comprises of about 1.5- 7 % of all oral pathoses.[5,6] It is a non -neoplastic vascular proliferation that is believed to be an exaggerated, conditioned response to a chronic low-grade irritation of the mucous membrane.[3]

These lesions can occur in all age groups but are mostly common in young females (1:1.5) in the second decade of life with 16% of cases associated with local etiologic factors.[7]

75% of oral pyogenic granulomas occur in the gingiva making it the most common site of occurrence. This could be due to the accumulation of calculus or other foreign material within the gingival crevice. Interestingly, these lesions are more common in the facial aspect of the anterior maxillary gingiva. However, occasionally some lesions have been reported to extend into the interproximal areas, involving the lingual and vestibular aspects of gingiva.[8] It can also occur on the lips, tongue and buccal mucosa.[3]

Literature search revealed very few cases of oral pyogenic granuloma associated with actinomycosis. One such case was reported by K Kuyama et al. on the maxillary gingiva, with

Fig. 1: Epithelium and connective tissue with encapsulating meshwork of basophilic structures. H and E (4X magnification)

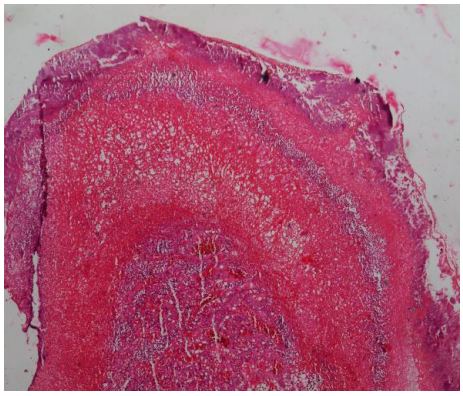

Fig. 2: Basophilic filamentous structures encapsulating the lesion in a palisading pattern (10Xmagnification)

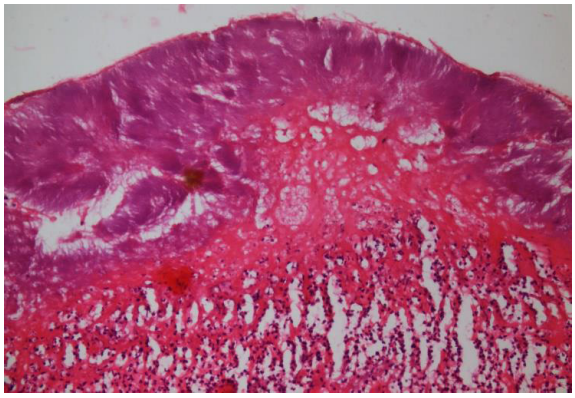

Fig. 3: Basophilic filamentous structures with peripheral club shaped ends. (40X magnification)

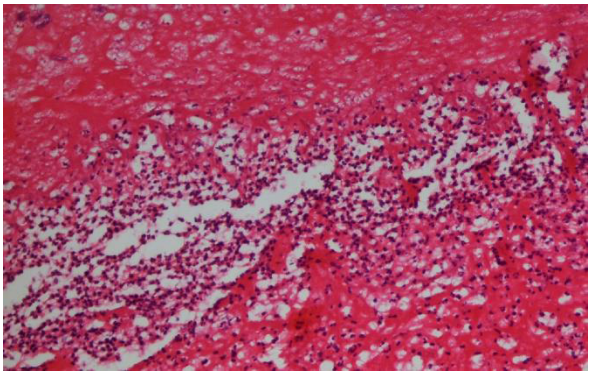

Fig. 4: Sea of neutrophils adjacent to the filamentous structures. (40X magnification)

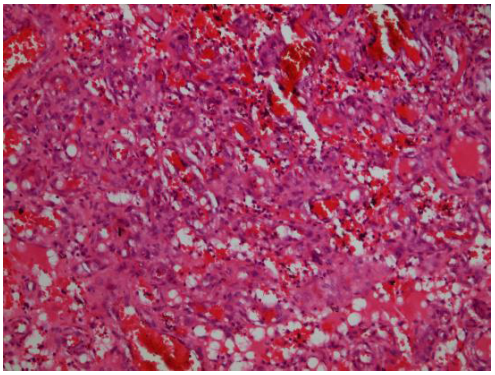

Fig. 5: Connective tissue showing mixed inflammatory cells and budding endothelial capillaries (40X magnification)

clinical and histological features corroborating with the present case.[9] However, few other cases were documented at sites distant from the oral cavity.[10] To the best of our knowledge, the current case describes pyogenic granuloma with actinomycosis in a site that has never been reported before in literature, making it an unusual one.

Varied etiopathogenesis have been attributed to the causation of this lesion such as local irritation, minor trauma, sex hormones, certain drugs like cyclosporine, defective restoration, calculus and poor oral hygiene. However, it is now widely agreed upon that pyogenic granuloma mainly arises from minor traumatic injury to the oral tissues though it may not always be evident in the clinical history of the patient. Although pyogenic granuloma was originally believed to be caused by pyogenic organisms, it is now considered to be unrelated to infection. Contrary to this view, some authors have associated infectious organisms to pyogenic granulomas, especially the recurrent ones.[8]

Actinomycosis is a chronic suppurative granulomatous bacterial infection caused by Actinomyces israelli. This rare disease is characterised by a slow, progressive endogenous spread and formation of multiple abscesses and sinus tracts with possible drainage of yellow "sulfur granules". It can also manifest as a soft tissue swelling or ulcer and is often misdiagnosed to be a neoplasm.[11] It shows a slight male predominance with a peak incidence in the 4th to 6th decades of life with predilection for immunocompromised individuals.[4] The patient in the present case was also in similar age range, but in contrast, she was a female.

Actinomyces species are gram positive, non–acid-fast, anaerobic or facultative anaerobic bacteria which are normal commensal flora of the human oral cavity. Though it is not considered as an invasive or opportunistic bacterium, it certainly becomes invasive on gaining entry to the submucosal tissues. Disruption of the mucosal barrier due to trauma to the oral tissues may serve as a portal of entry to these microorganisms.[11] Periodontal pockets, tooth extraction, poor dental hygiene, maxillofacial trauma or immunodeficiency can act as predisposing factors for actinomycosis.

The patient in our report however, could neither recall the time or situation of the initiation of the lesion nor did she give any history of trauma in the affected area. The biologic principle of tissue reaction emphasizes the fact that in an adult living tissue, any irritant applied can either act as a stimulus or a destructive agent or both.[3] It is interesting to note that pyogenic granuloma exhibits a hyperactive response to a stimulus for increased proliferation of vasculature and budding of endothelial cells in its collagenous connective tissue. Naturally, that makes it an aerobic environment which should ideally be non-conducive for thriving of the actinomyces species due to their anaerobic or microaerophilic nature. Therefore, the origin of the intriguing question, what was the initial disease?

This may be explained as follows. An initial actinomycosis infection may have been present in the affected site owing to either patient's poor oral hygiene or some mucosal breach in the buccal mucosa. This persistent presence of actinomycosis infection may have served as a chronic irritation (stimulus), followed by proliferation and budding of capillaries due to the upregulation of vascular morphogenetic factors leading to the formation of pyogenic granuloma in the same site.

Another situation could have been the initiation of an exaggerated inflammatory response due to a stimulus leading to increased angiogenesis and formation of pyogenic granuloma with actinomycosis presenting as a superimposed infection. The actinomyces species may have penetrated the submucosal tissue through a break in the mucosa in the site with existing pyogenic granuloma. However, the mechanism of their growth in an aerobic environment remains unclear as the culture of these organisms are extremely difficult. Although it is fair to say that the oral cavity exhibits pathogenic synergy where both aerobic as well as anaerobic organisms co-exist and even facilitates the growth of one another.[12]

Clinically pyogenic granuloma appears as deep red, smooth or lobulated soft exophytic mass which may be sessile or pedunculated and usually asymptomatic. It should be differentiated from other similar looking lesions of the oral cavity such as fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma, hemangioma.[3,8] Actinomycosis should also be included in the differential diagnosis of pyogenic granuloma and of any homogenous soft tissue lesion associated with inflammatory reaction due to its deceptive clinical appearances.[11]

Histologically, pyogenic granuloma has two types: lobular capillary hemangioma (LCH) and non-LCH. The LCH type has proliferating blood vessels organized in lobular aggregates with no specific changes such as edema, capillary dilation or inflammatory granulation. The non-LCH type, on the other hand, consists of a vascular core resembling granulation tissue with foci of fibrous tissue.[9, 13] The reported case resembled the LCH type.

Although it is extremely difficult to culture the organisms, prolonged bacterial culture in anaerobic conditions, special stains such as Methanamine silver and PCR techniques may be useful for identification of actinomyces species but histopathology remains the gold standard method. [6,8]

Conclusion

Only actinomycosis was detected in the present case alongside pyogenic granuloma, however the possibility of infection with other organisms cannot be eliminated. Whether the two lesions appeared simultaneously or one preceded the other remains a mystery and requires further exploration to uncover their nature. Clinicians should thus be aware about the occurrence of such rare combination of intraoral lesions, in order to make an accurate diagnosis and in turn, tailor a correct treatment protocol for these patients.

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