Case Report - Surgical Pathology

Oral pulse granuloma associated with keratocystic odontogenic tumor: Report of a case and review on etiopathogenesis

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ABSTRACT

Pulse granuloma is a distinct oral entity characterized as a foreign body reaction occurring either centrally or peripherally. It is usually seen in the periapical or in the sulcus area. Occasionally the lesions occur in the wall of the cyst, commonest being the inflammatory odontogenic cyst. Histologically, they present as eosinophilic hyaline mass with giant cell inclusions and inflammatory cells. They may show different histological characteristics, possibly related to the length of time in the tissue. Adequate recognition is important to avoid misdiagnosis. Many authors suggest that pulse granuloma results due to implantation of food particles of plant or vegetable origin into the tissue following tooth extraction. This paper aims to report a case of pulse granuloma associated with keratocystic odontogenic tumor with its histochemical and polarizing microscopic features and discuss on etiopathogenesis of pulse granuloma.

Keywords: Keratocystic odontogenic tumor, mandible, odontogenic keratocyst, pulse granuloma

INTRODUCTION

Pulse granuloma (PG) is an uncommonly reported oral finding having controversial etiopathogenesis, as many authors consider vegetable origin although others deny this.[1-4] In literature it has been described under various terminologies; like chronic periosteitis, granuloma in edentulous jaws, giant cell hyaline angiopathy, etc.[5-8]

Lewars in 1971 first described six cases of foreign body reaction in the mandible and thought it to be in response to food particles driven into the mucosa by a denture.[6] Subsequently, Adkins reported 15 cases with histopathologic features consisting of rings of palely stained structure less material and multinucleated giant cells around and within the rings. He suggested that they represent foreign bodies that might be the unresorbed residue of therapeutic agents inserted into extraction sockets and used the term granuloma in edentulous jaws.[7] In 1975, Rannie described these kinds of lesions as "chronic mandibular periostitis associated with vasculitis". Dunlap and Barker (1977)[8] used the terminology giant cell hyaline angiopathy and King (1978) first used the term pulse granuloma.[5] The term PG describes a granulomatous response, especially in lungs of infants or severely debilitated persons, after the aspiration of fragments of pulses.[5,9-12] Similar lesions have been found in the walls of the stomach.[5,12]

Investigators considered that the hyaline rings were residues of substances such as beans and peas.[13,14] However, few investigators did not endorse this view and suggested that hyaline rings instead represented degenerated blood vessels, degenerated collagen,[8] or fibrosed extravasated serum proteins.[2] Some even considered the lesion to be due to infection by Torulopsis glabrata.[15]

PG has rarely been documented in the walls of odontogenic cyst and there are few reported cases occurring in dentigerous cyst,[2,16] residual cyst,[2] and nasopalatine cyst.[8,17] The nature of these granulomatous lesions remains obscure. The present article

reports a case of PG in the wall of keratocystic odontogenic tumor (odontogenic keratocyst) and histochemical and polarizing microscopic appearance with emphasis on origin and pathogenesis of pulse granuloma.

CASE REPORT

A 56-year-old female patient reported with pain and difficulty in chewing food in the mandibular posterior region since one and a half months. Intraoral examination revealed bony hard swelling extending from alveolar ridge of mandibular first premolar to third molar with obliteration of vestibule. The mucosa over the swelling was normal. Right submandibular lymphnodes were enlarged [Figure 1a]. Patient gave past history of similar swelling eight months ago in the same region, for which she consulted a general dental practitioner. The right mandibular 1st and 2nd molars were extracted and enucleation of the cyst was done by the practitioner. Later patient did not report back to the practitioner as healing was uneventful and she was unaware of the previous diagnosis. The swelling reoccurred in the same area after four weeks. The alveolar mucosa appeared apparently normal with no change in color.

Orthopantomograph revealed a well defined radiolucency involving right mandibular body extending from mandibular first premolar to third molar region distally [Figure 1b]. A differential diagnosis of odontogenic keratocyst and ameloblastoma was considered and incisional biopsy was done.

Microscopic examination revealed a cystic epithelium overlying an inflammatory connective tissue. The epithelium was parakeratinised stratified squamous type with palisaded cuboidal basal cells. The epithelium demonstrated separation from underlying connective tissue in few areas [Figure 2a]. An interesting finding was presence of homogenous, pale, eosinophilic material resembling vegetable matter was seen dispersed in the cyst wall. It demonstrated a double layered refractile hyaline membrane surrounded with prominent chronic inflammatory cells and few foreign body giant cells [Figure 2b].

A diagnosis of keratocystic odontogenic tumor with pulse granuloma was made. The lesion was subsequently excised and application of Carnoy's solution was done with no evidence of recurrence since six months. The excised specimen confirmed the diagnosis of keratocystic odontogenic tumor and no vegetable matter was observed.

Histochemical staining

Histochemical staining with Periodic acid Schiff (PAS) without diastase, van Gieson and Alcian blue at pH 2.5 were performed. PAS [Figure 3a] and alcian blue [Figure 3b] demonstrated positivity for hyaline material confirming it to be vegetable matter. The surrounding condensed collagen stained reddish in color with van Gieson suggestive of matured collagen [Figure 3c].

Polarised microscopic findings

Vegetable matter presented as birefringent refractile material

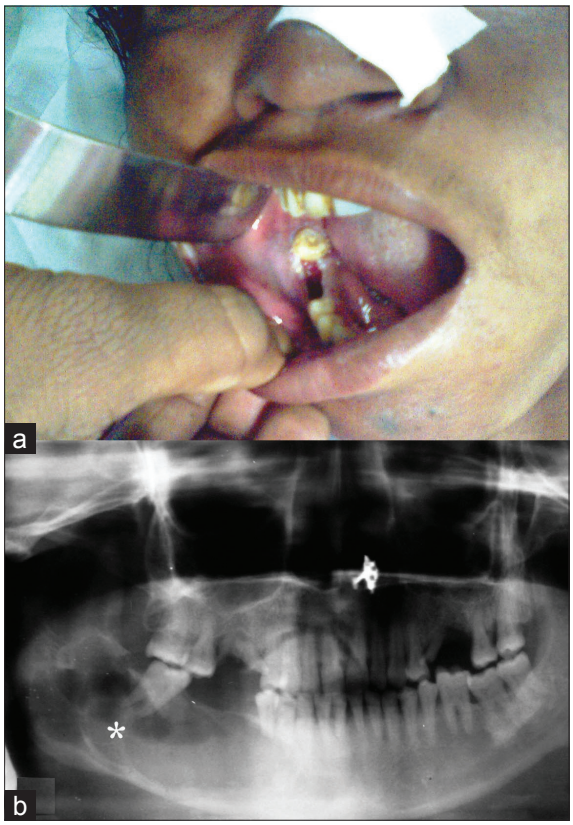

Figure 1: (a) Intraoral photograph showing cystic cavity after surgical exposure. (b) Orthopantomograph showing well demarcated radiolucent lesion extending from mandibular first premolar to third molar.

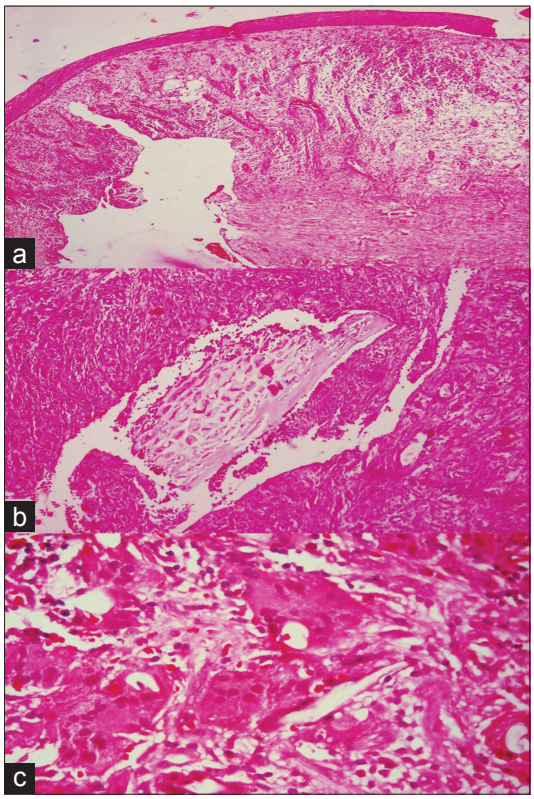

Figure 2: (a) Photomicrograph showing cystic cavity of odontogenic keratocyst (H and E, ×100). (b) Photomicrograph showing eosinophilic vegetable matter in cyst wall (H and E, ×100). (c) Photomicrograph showing foreign body giant cells (H and E, x40)

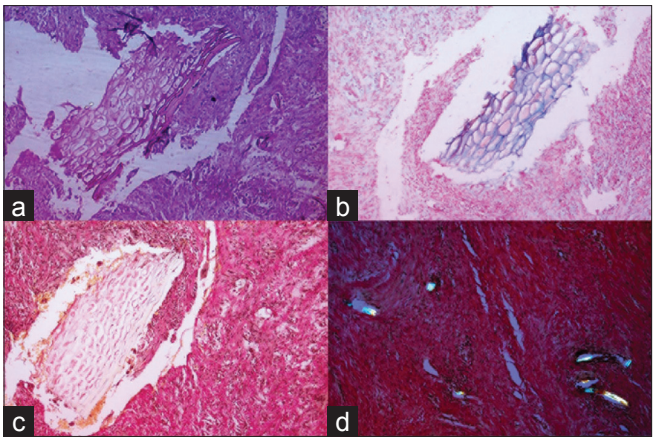

Figure 3: (a) Photomicrograph showing vegetable matter demonstrating PAS positivity (PAS, 100x), (b) Photomicrograph showing vegetable matter demonstrating alcian blue positivity (Alcian blue, x100), (c) Photomicrograph showing vegetable matter demonstrating van Gieson negativity (van Gieson, 100x). (d) Photomicrograph showing vegetable matter demonstrating vegetable matter demonstrating birefringence under polarized microscope. (100x)

under polarized microscopy [Figure 3d].

DISCUSSION

Pulse granuloma is an uncommonly reported oral finding with undefined etiology.[1] They appear as spherical, ovoid, or irregular bodies surrounded by fibroblasts and appear as homogeneous, eosinophilic material in Hematoxylin and Eosin stained section surrounded by acute and chronic inflammatory cells and foreign body giant cells. Occasionally, there may be presence of small round calcified basophilic bodies within the amorphous hyaline material.[3]

Many authors believe this hyaline material to be a vegetable matter embedded within the edentulous portion of jaw bones, periapical lesions, and extraction sockets or in the wall of the odontogenic cysts, etc. Controversy regarding the terminology for this lesion has been a subject of discussion since its inception. King used the term oral pulse granuloma, for lesions occurring in the oral cavity.[5] In the literature, a total of 170 cases of oral and extra oral cases of pulse granuloma have been reported.[18] Out of these 41 cases have been reported in the cystic walls and among these 33 cases are associated with inflammatory odontogenic cyst[2,3,5,13,19-21] and only eight cases are associated with odontogenic cyst, out of this two cases of dentigerous cyst[2,16] three cases of nasopalatine cyst[3,8,17] and three cases of keratocystic odontogenic tumor (odontogenic keratocyst) have been reported.[3] Thus, occurrence of pulse granuloma in wall of odontogenic cyst is a rarity. We report one such rare case of pulse granuloma in the wall of keratocystic odontogenic tumor.

Chou *et al*., have described these lesions as a distinct entity and classified them into central and peripheral according to the location. Central lesions are asymptomatic and peripheral lesions presents as painless submucosal swellings. The pathogenesis of pulse granuloma has been defined as exogenous or endogenous in origin. The concept of exogenous origin is based on the implantation of food particles of plant origin, through extraction sockets, deep periodontal pockets, infilled root canals and grossly decayed teeth.[5,21,22] Talacko and Radden hypothesized that once the food gains access to tissues, it is rapidly digested and altered by the host responses. The cellulose moiety of plant foods is indigestible and persists in the form of hyaline material, inciting a chronic granulomatous response.[3]

Adkins considered the lesion to be a foreign body granuloma, with the hyaline material being the foreign body and the origin of the foreign body was unknown[5,7] Rannie, Mincer, and co-workers considered that the foreign body reaction is due to legume cells, which contain starch and they termed it as pulse granuloma[14,17,23] Levison *et al*., in their study, revealed the presence of an unexpectedly wide range of elements in routine biopsies, indicating that implantation can occur without gross trauma. Pulse granuloma is commonly found in the mandibular posterior region where food stagnation is more common.[3,24] Harrison and Martin supported the theory of a vegetable nature of the hyaline rings on the basis of ultrastructural investigation. They presented evidence suggesting that the hyaline rings were mainly composed of cellulose, thus the term "oral vegetable granuloma" was proposed.[4,13]

The suggested endogenous origin is believed to be due to localized hyaline degenerative change in the walls of the blood vessels as given by Dunlap and Baker and they termed the condition as giant cell hyaline angiopathy.[8,13] Chen and his colleagues studied hyaline bodies in the connective tissue wall of odontogenic cysts and proposed three stage process for the hyaline ring formation, i.e., pooling and coagulation of extravasated serum proteins in areas of trauma, ring arrangement around the coagulum of protein and soldification of the hyaline body thus causing the entrapment.[1,5] Hase and coworkers reported an interesting case of *Torulopsis glabrata* infection in the oral cavity which presented clinically and histologically similar to pulse granuloma and considered pulse granuloma could be due to infection by *T. glabrata*.[15]

Based on histochemical and immunohistochemical analyses, Luiz Alcino Gueiros concluded that oral lesions are caused by traumatic implantation of vegetable particles in an extraction socket or oral ulcer with cellulose being responsible for granuloma formation.[1] The present case being that of recurrence implicates that the implantation of food particle might have occurred during the healing phase of cyst removal.

Radiographically, intraosseous pulse granuloma is an irregular radiolucent lesion with well formed trabeculae of bone, an appearance that can be confused with that of other jaw lesions. Extraosseous pulse granuloma presents as poorly defined erosion of the crest of the alveolar ridge, which may be similar to that of peripheral giant cell granuloma. Pulse granuloma is rarely diagnosed clinically and it remains largely a microscopic diagnosis. The infrequency of this diagnosis may be attributed to a sampling phenomenon. If more tissue sections of cases involving clinical factors with a known association with pulse granuloma were prepared, the frequency of detection might increase. Another reason for under diagnosis may be that the tissue response to particular vegetable matter may be trivial and in most cases it does not lead to gross clinical lesion.[13] Treatment for pulse granuloma

is complete surgical excision of the lesion. The recurrence of pulse granuloma is rare and if recurs it is probably due to incomplete excision.[5] In the present case, complete surgical excision of the lesion with carnoy's solution application was done.

Histologically, the oral pulse granuloma contains starch granules, with cellulose envelopes that appear as hyaline rings surrounded by foreign body giant cells and concentrically arranged delicate connective tissue.[5,9,10] Luiz Akino *et al*., studied three cases of pulse granuloma and found that more giant cells were seen in initial lesions then in older ones, which showed droplet calcifications within eosinophilic masses. Hyaline rings in the present case stained strongly for PAS as demonstrated by various authors.[1,3,4] They demonstrate definite birefringence under polarized light,[3,22] which was evident even in our case. Additionally, alcian blue showed positivity for vegetable material. Vegetable cell walls contain acidic groups such as carboxyls which are stained by alcian blue, while van Gieson was negative, which was in accordance with other authors.[3,4]

In conclusion, pulse granuloma is a well described entity with distinct histopathology. Occurrence of pulse granuloma can be well documented if extensive sampling is done. Oral vegetable implantation has distinct histopathological aspects from pulse granulomas of the lungs and gut as starch cells are often absent and giant cells may be scant. Once it promotes and maintains chronic inflammation, however, it can be responsible for a distinct and persistent histological evolution, especially in an intraosseous location. This case report was presented to emphasize the occurrence of pulse granuloma in recurrent lesion, which is mainly due to implantation of food particle in a healing wound. Thus, postoperative maintenance of oral hygiene is very important to avoid embedding of vegetable matter into healing wound.

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