

# Oral pulse granuloma associated with ameloblastoma: Report of a case and review of literature

Uma Vasant Datar, Rahul B Patil<sup>1</sup>, Aarti M Mahajan<sup>1</sup>, Sachin Dahivelkar<sup>2</sup>

Department of Oral Pathology and Microbiology, Bharati Vidyapeeth Deemed University Dental College and Hospital, Sangli, Departments of <sup>1</sup>Oral Pathology and Microbiology and <sup>2</sup>Oral Surgery, MGV's Dental College, Nashik, Maharashtra, India

## Abstract

Oral pulse granuloma (PG) is a rare lesion that presents as a foreign body granulomatous reaction to implanted food particles of plant or vegetable origin. It is classified as a central or a peripheral lesion depending on the location. Rarely, a central oral PG may be associated with a cyst or tumor, thus leading to a deceptive histopathological picture. This is exemplified in the present case of 27-year-old female patient who reported with a swelling on the left posterior region of mandible which was diagnosed as ameloblastoma. The excisional specimen depicted a varied histopathological picture showing follicular ameloblastoma and foreign body granulomas associated with hyaline rings. Based on histopathological, histochemical and polarized microscopy findings; these hyaline rings were diagnosed to be remnants of a plant cell/legume. The aim of this article was to present a case of PG associated with ameloblastoma and discuss its histochemical and polarizing microscopic features.

**Keywords:** Follicular ameloblastoma, foreign body granuloma, oral pulse granuloma, oral vegetable granuloma

## Address for correspondence:

Dr. Uma Vasant Datar, Department of Oral Pathology and Microbiology, Bharati Vidyapeeth Deemed University Dental College and Hospital, Sangli - 416 415, Maharashtra, India. E-mail: dataruv@gmail.com

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## INTRODUCTION

Oral pulse granuloma (PG) was reported by Lewars in 1971.<sup>[1]</sup> Endogenous theory<sup>[2,3]</sup> and exogenous theory<sup>[4-7]</sup> have been put forth to explain the etiology of PG. The exogenous theory is most accepted, and the lesion is termed as “pulse granuloma” since it is a foreign body reaction to entrapped leguminous food/pulses.<sup>[5,6]</sup> Microscopically, PG is characterized by the presence of giant cells and hyaline rings.<sup>[6]</sup> PG has been reported in the walls of odontogenic cysts.<sup>[5,7-10]</sup> Only one case has been reported till date of PG associated with ameloblastoma.<sup>[9]</sup> Herein, we report a

case of PG associated with ameloblastoma and discuss its etiopathogenesis, polarizing microscopy and histochemical findings.

## CASE REPORT

A 27-year-old female patient presented with a history of swelling in mandibular left posterior region for 1 year and pain for 1 month [Figure 1]. Intraoral examination revealed a bony hard swelling extending from the left mandibular second premolar till retromolar area. Obliteration of buccal and lingual vestibule was noted. The mucosa overlying

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the lesion appeared normal. The patient had undergone extraction of mandibular first molar of the same side earlier.

The radiographic examination revealed a multilocular radiolucency involving left mandibular region [Figure 2a and b]. The lesion extended from the second premolar and posteriorly it involved the coronoid and the condylar processes. The expansion of buccal and lingual plate of the mandible was evident on occlusal radiographic images. Differential diagnoses of ameloblastoma and keratocystic odontogenic tumor were considered. Incisional biopsy confirmed the diagnosis of follicular ameloblastoma. Based on the histopathological diagnosis, segmental block resection was carried out with the surgical margin 1 cm away from the radiographic boundary of the lesion. The excised specimen was sent for histopathological diagnosis. No evidence of recurrence was noted after 1 year of resection.

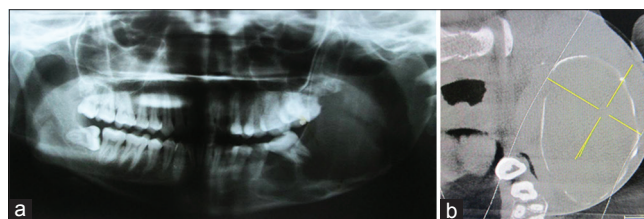
The gross specimen showed expansion of buccal and lingual plates of mandible and bone erosion in the retromolar area. On grossing, a large cystic lesion was noted involving body and ramus of mandible. Microscopic examination of the excised tissue specimen revealed follicles of ameloblastoma of varying sizes [Figure 3a]. The stroma was mature, collagenous and it demonstrated foreign body granulomas with numerous multinucleated giant cells [Figure 3b and c]. On careful examination, it was evident that these foreign body granulomas were associated with multiple, amorphous, eosinophilic masses enclosed in densely hyalinized eosinophilic matrix/rings [Figure 3b and c].

Histochemical staining was done to understand the nature of hyaline rings. The hyaline rings were periodic acid–Schiff positive [Figure 3d]. The peripheral portion of the foreign body was positive for Masson's trichrome stain suggesting it to be condensation of collagen.

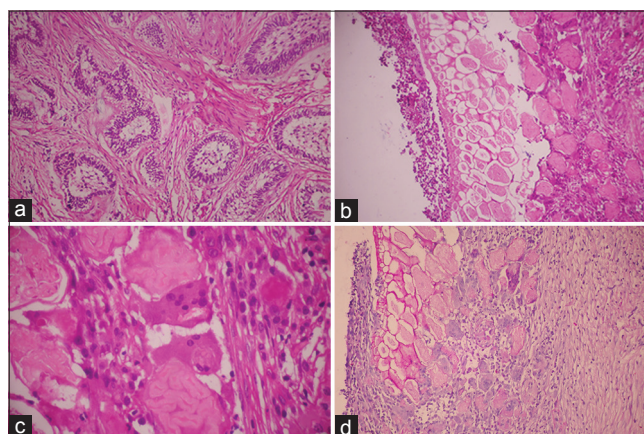
To prove the similarity of the foreign body and plant cells, grains such as gram, wheat, rice, split pigeon pea were boiled and processed. Moreover, vegetables such as carrot, cabbage were processed raw. The sections were stained with hematoxylin and eosin and periodic acid-Schiff. We observed that a section of a legume [Figure 4b] bore a striking resemblance, to the foreign body we encountered. Both the legume and the foreign body showed peripherally smaller angular to rectangular cells and centrally larger cells enclosing amorphous substance. When viewed under polarized light, the hyaline structures exhibited birefringence [Figure 5a and b] and the fragments of



**Figure 1:** Clinical photograph shows a diffuse solitary swelling on the left side of the face, involving the angle of mandible



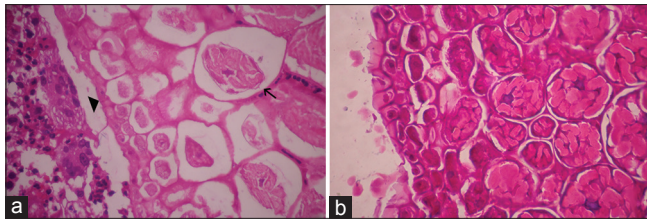
**Figure 2:** (a) Orthopantomograph showing multilocular radiolucency in the mandibular body-ramus area on the left side. (b) Cone beam computed tomography image showing buccal expansion due to the lesion



**Figure 3:** (a) Photomicrograph demonstrating follicles of ameloblastoma in mature fibrous stroma (H&E, ×10). (b) Foreign body and associated granulomas (H&E, ×10). (c) Multinucleated giant cells phagocytosing hyaline ring-like foreign particles (H&E, ×40). (d) The foreign body demonstrating periodic acid–Schiff positivity (periodic acid–Schiff, ×10)

material similar to hyaline rings were noted within the giant cells [Figure 5c and d].

Based on these observations, the foreign bodies were identified as remnants of food particles of vegetable origin. To rule out other local and systemic



**Figure 4:** (a) Foreign body showing peripheral smaller rectangular cells (demonstrated by arrowhead) and arrow pointing larger more angular cells in the center enclosing amorphous eosinophilic material (H&E, x40). (b) Processed pulse showing structure similar to the foreign body (H&E, x40)

granulomatosis, hematological investigations were done, and the results were within normal limits. Tuberculosis was ruled out by normal findings of chest radiography and negative tuberculin test. Hence, the lesion was diagnosed as follicular ameloblastoma with PG. Patient's consent has been obtained for publishing clinical findings and images.

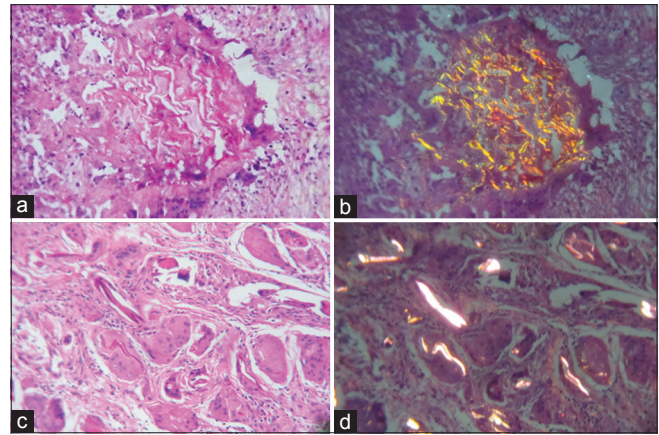
## DISCUSSION

PG is a rare granulomatous lesion characterized with so-called hyaline ring structures which are always associated with multinucleated giant cells.<sup>[4]</sup> PG is known by many terminologies such as chronic periostitis,<sup>[1]</sup> giant cell hyaline angiopathy,<sup>[2]</sup> hyaline ring granuloma,<sup>[3]</sup> vegetable granuloma<sup>[4]</sup> and oral pulse/hyaline ring granuloma.<sup>[5]</sup>

According to a recent review by Philipsen and Reichart,<sup>[5]</sup> 173 cases of PG have been reported till date. In oral and maxillofacial region, these lesions often are noted in association with odontogenic and nonodontogenic cysts, but very rarely in odontogenic tumors.<sup>[5,7-12]</sup> Talacko and Radden<sup>[7]</sup> in their retrospective study, found evidence of PG in periapical lesions and odontogenic cysts which were missed initially at the time of diagnosis. Similarly, Henriques *et al.*<sup>[10]</sup> retrospectively studied inflammatory odontogenic cysts and found that 3.3% (22 of 661 cases) of inflammatory cysts were associated with PG. In the majority of cases, the response to foreign body is minimal; hence, it can be missed and which may be reason for underreporting of PG.<sup>[11]</sup> Thorough literature search revealed that only one case of PG has been reported in association with ameloblastoma.<sup>[9]</sup> This may be because the prevalence of ameloblastoma is low, and histopathologists are unfamiliar with PG; hence, such a lesion may have been overlooked in the past.

Over the years, etiopathogenesis of PG has invited a lot of controversies. Two paradoxical theories have been proposed to explain the etiopathogenesis of PG.

1. The endogenous theory states that the hyaline rings



**Figure 5:** (a and b) Photomicrographs showing the features of irregular foreign body particles in light microscopy and polarizing microscopy, respectively (H&E, x40). (c and d) Foreign body giant cells with ingested material which shows birefringence and hence is evident on polarizing microscopy (H&E, x40)

originate due to degenerative change in blood vessels<sup>[2]</sup> or due to extravasated serum proteins<sup>[3]</sup>

2. The exogenous theory states that PG is a foreign body granuloma, which is induced by the presence of food particles of vegetable/plant origin.

Harrison and Martin<sup>[4]</sup> proved the vegetable nature of hyaline rings based on ultrastructure findings. Talacko and Radden<sup>[6]</sup> through animal model have suggested that PG results from traumatic implantation of food particle of plant origin. The starch in the plant cell gets digested easily leaving behind cellulose which is undigestible and it invokes a granulomatous response. The cellulose of legumes is more noxious than other vegetable cells because of its resistance to digestion by phagocytes. They also have suggested that legumes are rich in phytohemagglutinin, which are known to have granuloma enhancing properties.<sup>[6]</sup>

PG may present as a central lesion or a peripheral lesion. In case of the central lesions, the theory of exogenous origin is based on the premise that the implantation of vegetable particles occurs through extraction sockets, pericoronal flap, grossly decayed teeth, deep periodontal pockets, unfilled canals and through a pathological communication with oral cavity.<sup>[8,9]</sup> In the present case, perforation of cortical plates was noted in the retromolar area which could have helped in entry of vegetable matter.

Microscopically, PG is characterized by aggregates of thick eosinophilic hyaline matrix or hyaline rings surrounded by chronic inflammatory cells and multinucleated giant cells. The number of giant cells is variable, and the stroma may or may not be fibrosed. These hyaline rings are



of varying size, shape and thickness. The hyaline rings often enclose starch.<sup>[4]</sup> This characteristic arrangement is reminiscent of vegetable starch cell.<sup>[5]</sup> A legume (pulse) has an outer shell surrounding a cotyledon. The cotyledon has a honeycomb structure within which stored food, i.e., starch is noted.<sup>[12]</sup> In the present case, the foreign body was composed of vertically oriented stratified cells. The cells at periphery were rectangular, joined end to end; each cell had a well-defined cell wall. The outer rectangular cells demonstrate thick outer wall which indicates it to be a part of seed coat [Figure 4a]. Thus, the histology of foreign body in present case bore a striking resemblance to a cooked legume [Figure 4b]. Hence, we are of the opinion that the term “oral PG” is more appropriate for this lesion.

The hyaline rings have been reported to be birefringent under polarized light<sup>[6,8]</sup> which was evident in the present case. Numerous giant cells were seen to contain ingested birefringent material [Figure 5b and d].

The chronic granulomatous reaction triggered by cellulose moiety of PG may be confused with other granulomatous lesions such as tuberculosis, Wegener's granulomatosis, midline lethal granuloma, Crohn's disease, allergic oral granulomatosis, Melkersson–Rosenthal syndrome and sarcoidosis to name a few.<sup>[9]</sup> In the present case, erythrocyte sedimentation rate was not raised, hemoglobin levels, differential white blood cell, total white blood cell and red blood cell counts were within normal limits. The systemic granulomatosis was ruled out based on the absence of clinical signs and negative hematological investigations.<sup>[9]</sup>

According to Philipsen and Reichart,<sup>[5]</sup> for diagnosis of PG, the following histopathological features are essential:

- Eosinophilic hyaline rings in the form of circular homogenous or fibrillar masses
- A chronic inflammatory lesion
- Foreign body giant cells.

Thus, the histopathological, histochemical, polarizing microscopy findings and the observations of Philipsen and Reichart<sup>[5]</sup> lead to the final diagnosis of PG. Adequate tissue sampling, serial sectioning, use of special stains and microscopy techniques may be beneficial to establish the diagnosis of PG. The treatment of PG is complete excision and it does not recur after excision.<sup>[8]</sup>

## CONCLUSION

We report a rare case of PG associated with follicular ameloblastoma and highlight the diagnostic features of PG. The present case emphasizes that histopathologists should be acquainted with PG to avoid misdiagnosis, especially when it occurs in conjunction with other discrete histopathologic entity.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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