Unicystic Ameloblastoma with Pulse Granuloma: A Case Report and Literature Review with an Emphasis on Pathogenetic Mechanisms

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Abstract

Innate immunity is essential for detecting and eliminating foreign substances, and oral pulse granuloma (OPG) is a rare reaction to vegetable or pulse material often seen in the oral cavity. This report presents a unique case where a dentigerous cyst transformed into unicystic ameloblastoma with concurrent OPG, offering insights into its development. A 15-year-old boy came to us with swelling in the right lower jaw. Clinical and X-ray findings pointed to an impacted wisdom tooth, and a dentigerous cyst was suspected. However, after surgical removal and closer examination under the microscope, the cyst was unexpectedly diagnosed as unicystic ameloblastoma containing unusual hyaline bodies. Further testing confirmed these as vegetable matter, leading to a diagnosis of OPG. The case displayed a rare "hyaline predominant" phase of OPG, fitting into stages of a foreign body reaction: material impaction, inflammation, giant cell response, and scarring. This case underscores the importance of detailed tissue analysis and special staining to identify OPG and differentiate it from other similar conditions. The findings highlight the rare coexistence of OPG with a cyst transforming into ameloblastoma, emphasizing the need for careful diagnosis to guide effective treatment.

Keywords: *Foreign body, giant cells, granulomatous reactions, odontogenic*

Introduction

Innate immunity plays a pivotal role in identifying and eliminating infectious agents and foreign materials. The ability to identify self and nonself-antigens by the innate immune cells helps to turn down the potential threat posed by noxious antigens. The foreign body can be eliminated by phagolysosomal digestion into nondangerous, reusable, and excretable subunits.[1] A well-coordinated regulatory mechanism may fail at times and result in a granulomatous inflammation culminating in granuloma formation. Oral pulse granuloma (OPG) is a granulomatous inflammatory lesion arising as a result of a foreign body reaction (FBR) to vegetable/pulse material. OPG is encountered commonly in the oral cavity in association with an array of pathologies such as periapical granuloma, odontogenic cysts, nonodontogenic cysts, odontogenic tumors, osteomyelitis, and many others.[2] We describe a case of a dentigerous cyst that developed into ameloblastoma in conjunction with a pulse granuloma, and we briefly explore the pathogenetic pathways for OPG in an effort to put out a theory that explains the gradual development of OPGs.

Case Report

A 15-year-old male patient reported to our institute with swelling in the right lower back region of the jaw for 3 months and pain for 15 days. Extraoral examination revealed a diffuse, nontender swelling in the right mandibular angle region of the jaw. Swelling extended from the right-side mid-body region of the mandible till the angle of the mandible. Intraoral examination showed a diffuse swelling obliterating the buccal vestibule at the right posterior region of the jaw. The swelling extended from 46 to pterygomandibular region. There was a clinically missing 48 with a pericoronal flap at the distal aspect of 47 [Figure 1]. OPG showed a well-defined unilocular expansile corticated radiolucent lesion in the right mandibular angle with impacted right third molar displaced inferiorly till the lower border of the mandible. The radiolucency was

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noted pericoronal to 47 and extended up to ramus of the mandible [Figure 2]. A provisional diagnosis of dentigerous cyst was given.

Fine-needle aspiration was done. It yielded straw-colored fluid. On wet mount examination, the findings were noncontributory with only chronic inflammatory cells being evident. An incisional biopsy was performed. On histopathologic examination, a cystic lumen lined by tall columnar cells with hyperchromatic nuclei showing reversal of polarity satisfying Vickers–Gorlin criteria which was suggestive of ameloblastomatous lining was noted. This lining at areas showed intraluminal proliferation [Figure 3]. The connective tissue was dense, fibrous, and haphazardly arranged with the absence of ameloblastic epithelium. Histopathological diagnosis of the intraluminal variant of unicystic ameloblastoma was made.

After establishing the microscopic diagnosis, surgical excision was done. Histopathological examination of the completely excised specimen revealed a cystic lumen lined by epithelium of 2–3 cell layer thickness. The cystic wall was loose myxomatous and at places fibrous. Foci of giant cells were also evident in the cystic wall surrounding numerous hyaline eosinophilic bodies with chronic inflammatory infiltrate [Figure 4]. The current microscopic picture was completely different from the earlier obtained postincisional biopsy. To confirm the origin of hyaline bodies special staining was done. The hyaline bodies were periodic acid–Schiff (PAS) positive and also revealed vegetable cell wall which was disguised on routine staining. Hyaline bodies were negative for Van *Gieson's stain.* Special stains confirmed that the hyaline bodies were a host response to pulse or vegetable material, marked by foreign body giant cell formation. Based on the incisional and excisional biopsy, a diagnosis of dentigerous cyst transforming into ameloblastoma with pulse granuloma was made.

During the 2nd-month follow-up, the patient showed signs of healing with respect to the lesional site.

Discussion

Pulse granuloma is a FBR to vegetables or pulses, caused by cellulose and starch in their cell wall. It is commonly

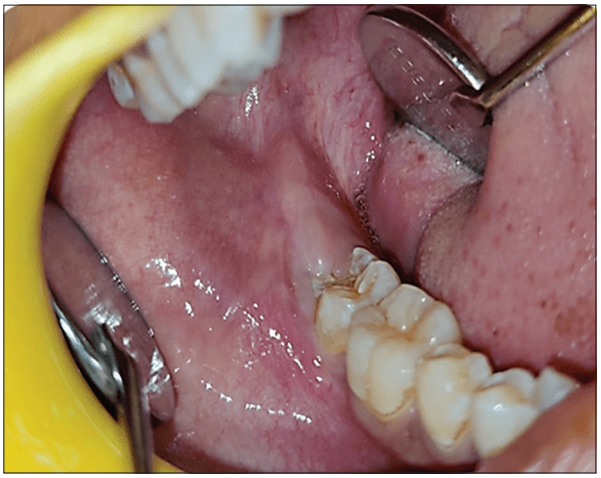

Figure 1: Gross view of the diffuse swelling obliterating the right lower buccal vestibule. Note the pericoronal flap covering the distal aspect of 47

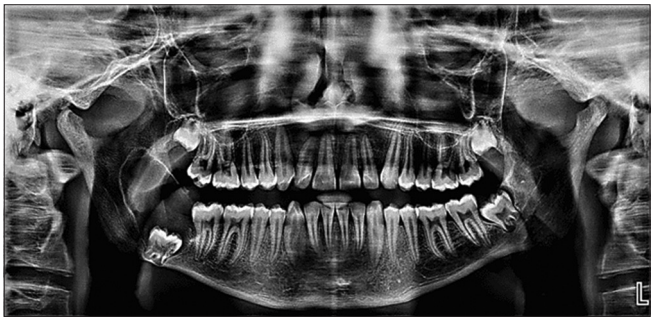

Figure 2: Orthopantomogram of the mandible exhibits a well-defined unilocular expansile corticated radiolucent lesion on the right side of the mandibular angle with impacted third molar displaced inferiorly

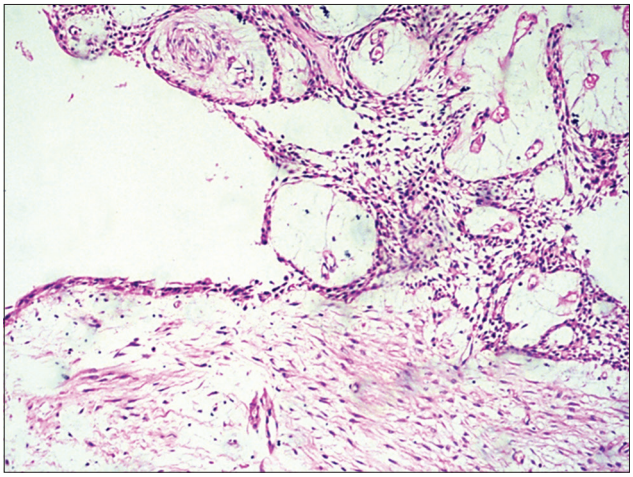

Figure 3: Unicystic ameloblastoma. Dentigerous cyst lining exhibiting focal area of ameloblastomatous changes with luminal proliferation of ameloblastomatous lining (H and E, ×10)

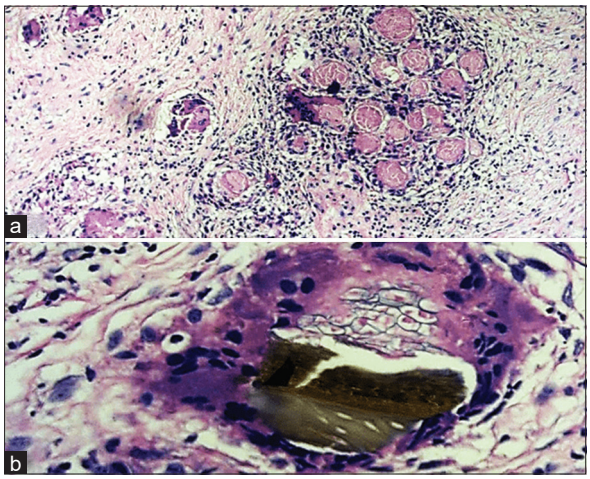

Figure 4: Photomicrograph of pulse granuloma. (a) Hyaline eosinophilic bodies with chronic inflammatory infiltrate, foreign body giant cells, and fibrous wall (H and E, ×10). (b) Foreign body giant cell exhibiting engulfed pulse material (H and E, ×40)

seen in the oral cavity,[2] sinonasal tissues,[3] lungs, and gastrointestinal tract.[4] OPG is usually clinically indolent and incidental but may form a mass lesion at times.

OPG in literature is described under various names such as hyaline ring granuloma, giant cell hyaline angiopathy, hyaline bodies, oral vegetable granuloma, vegetable induced granuloma, and chronic mandibular periostitis. OPG commonly occurs in edentulous alveolar ridges with a history of tooth extraction, the periapical region of teeth requiring root canal treatment, the open pulp chambers of decayed teeth, the walls of odontogenic cysts and tumors, and the pericoronal area of partially impacted third molars. In the present case, two possible conjectures can be drawn: first pulse material gaining access into the pericoronal pocket of 47 and embedding into the wall of underlying unicystic ameloblastoma and second an improper suturing postincisional biopsy or opening of suture acting as a portal entry for pulse material and implantation into the wall of unicystic ameloblastoma.

Numerous pathogenetic mechanisms are described in the literature. Dunlap and Baker have given the endogenous theory suggesting the hyaline degenerative changes in the walls of blood vessels caused by acute vasculitis. On the other hand, exogenous theory vocalizes the cellulose and starch moiety of pulse or vegetable incites FBR.[5] Some authors suggest *Torulopsis glabrata,* currently known as *Candida glabrata* as an etiology for pulse granuloma.[6] FBR is a sequential event. We put forth a possible hypothesis in FBR for OPG in six sequential stages [Table 1]:

i. Stage of impaction
ii. Stage of interface formation
iii. Stage of acute inflammation
iv. Stage of chronic inflammation
v. Stage of foreign body giant cell formation
vi. Stage of fibrosis.

Under routine staining, pulse granulomas appear as pale eosinophilic hyaline bodies that are round, oval, or irregular in shape. These are surrounded by acute and chronic inflammatory cells, foreign body giant cells, and, in mature lesions, a layer of fibrosis at the edges. Microscopically pulse granuloma of the mesentery is divided into three phases – cellular predominant, hyaline predominant, and sclerosing mesenteritis.[7] Similar phases can be described in the context of OPG. We propose three phases – cellular predominant, hyaline predominant, and sclerosing type. Cellular predominant exhibits abundance of acute and chronic inflammatory infiltrate around the hyaline material of pulse. The sclerosing type is characterized by extensive fibrosis surrounding the pulse material, while the hyaline-predominant phase shows minimal inflammatory cells and an abundance of hyaline material. The current case exhibited the hyaline predominant type.

Philipsen and Reichart have given diagnostic criteria[8] for pulse granuloma which should satisfy all three histopathological features:

1. Eosinophilic hyaline rings in the form of circular homogenous or fibrillar masses

Table 1: Hypothesis in foreign body reaction for oral pulse granuloma in six sequential stages

Stages	Events
Stage of impaction	Pulse material invades into the host tissue as a result of impaction. The impaction can occur as a traumatic event during mastication. Pulse material gains entry through gingival sulcus, open pulp chamber, or pathology such as cyst or tumor which alters the integrity of oral anatomy
Stage of interface formation	Implantation of pulse material is a traumatic event resulting into wound formation and local bleeding. Pulse material comes in contact with host blood. The serum proteins of host blood interact with surface of pulse material and after few hours fibrin dominated thrombus is formed at the interface of pulse material and host tissue
Stage of acute inflammation	This stage is characterized by infiltration of neutrophils and mast cells. A transitory phase lasting for few hours-days. This phase exhibits interaction of DAMPs expressed by traumatized host cells and PRRs expressed by neutrophils. Mast cells secrete interleukin-4 and 13. The elimination of the foreign body is not always possible at this stage and it soon culminates into chronic inflammatory stage
Stage of chronic inflammation	Chronic inflammation starts 2–5 weeks after implantation of vegetable material in oral tissue. Lymphocytes, macrophages, and plasma cells are predominant cells in this stage. Macrophages secrete IL-1, IL-6, IL-8, and TNF-α which boosts and continues the process of inflammation. Macrophages execute the process of eliminating pulse material by phagocytosis and by the release of ROS and lysosomal enzymes
Stage of FBGC formation	FBGC formation is the process of macrophage fusion which leads to large, up to several 100-μm-sized giant cells up to dozen of nuclei. The FBGC are of monocyte-macrophage lineage. IL-4 and IL-13 are responsible for fusion of macrophages which fail to digest the pulse material. FBGC is an example of higher differentiation with modified function to resist apoptosis and eliminate the pulse material
Stage of fibrosis	The end stage of FBR is formation of dense collagen fibers peripherally which walls the granuloma. Pro-fibrotic and angiogenic growth factors such as PDGF, VEGF, and TGF-β are responsible for formation of fibrous capsules. Proteolytic enzymes such as MMP secreted by macrophages, and endothelial cells are involved in the ECM remodeling

DAMPs=Damage-associated molecular patterns, PRRs=Pattern recognition receptors, IL-1=Interleukin-1, TNF-α=Tumor necrosis factor-α, FBGC=Foreign body giant cell, FBR=Foreign body reaction, PDGF=Platelet-derived growth factor, VEGF=Vascular endothelial growth factor, TGF-β=Transforming growth factor-beta, MMP=Matrix metalloproteinases, ECM=Extracellular matrix, ROS=Reactive oxygen species

2. A characteristic inflammatory lesion
3. Foreign body giant cells.

The present case exhibited all three histopathological traits.

Special stains that can be used to stain pulse material and hyaline bodies are PAS and Alcian blue stain owing to the starch moiety present in pulse or vegetable cell wall.[2] The pulse material is negative for Van Gieson. A similar observation was seen in the current case. Special stains like PAS can be of utmost importance to locate the pulse material which is otherwise difficult to locate in routine stains.

Pulse granuloma should be differentiated from other granulomatous diseases such as tuberculosis, sarcoidosis, and fungal and parasitic infections. Oral tuberculosis is extremely rare. Its histopathology typically shows caseating granulomas with filamentous bacilli, which can be identified using Ziehl–Neelsen staining. Sarcoidosis exhibits noncaseating granuloma with asteroid bodies in giant cells occasionally. Fungal infections of the oral cavity can be diagnosed on routine and special stains by assessing the morphology and stainability of fungal hyphae and spores. Parasitic infections of the oral cavity will show the presence of parasitic eggs or helminths in microscopy, and adjuvant serological tests can be done to confirm parasitic infection. Eosinophilic bodies encountered in oral lesions such as Rushton bodies, ghost cells, dentinoid material, and amyloid material should also be differentiated from hyaline rings of pulse granuloma. Rushton bodies are eosinophilic bodies seen in the epithelium of radicular cyst they are PAS negative. Ghost cells are enlarged epithelial cells with eosinophilic cytoplasm and absence of nuclei they are encountered in odontogenic cysts and tumors they are PAS negative. Immunohistochemical staining would reveal positivity for enamel proteins such as enamelin, sheathlin, enamelysin, amelogenin, and Barx protein. Dentinoid material is seen in odontogenic tumors of epithelium and ectomesenchyme origin as a result of inductive phenomenon. Dentinoid material stains green with modified Gallego's stain. To differentiate other FBRs a thorough history taking is important for example a surgeon using latex gloves with talc or *Lycopodium* dust during an incisional procedure of cyst or tumor which has evoked FBGR secondarily. When diagnosing a giant cell pathology, it's crucial for clinicians to take a detailed patient history, including information about any suture materials or implants that may have triggered a foreign body granuloma reaction (FBGR), as these factors can influence the diagnosis.

Conclusion

OPG is a distinct histopathological entity that is commonly encountered secondary to an array of oral lesions. It should be differentiated from other granulomatous diseases of oral and systemic origins. OPG may exhibit exuberant growth potential and impair healing postsurgery. There is no recurrence of OPG, but a pathologist should understand the pathogenesis and promptly diagnose it.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parent has given consent for images and other clinical information to be reported in the journal. The parent understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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