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IP Indian Journal of Anatomy and Surgery of Head, Neck and Brain

Journal homepage: <https://www.ijashnb.org/>

Case Report

Unusual case of hyaline ring granuloma associated with pericoronitis: Case report and review of literature

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ARTICLE INFO

Article history:

Received 27-05-2022

Accepted 16-06-2022

Available online 06-07-2022

Keywords:

Hyaline rings

Granulomas

Masson's

ABSTRACT

Objective: Oral Hyaline Ring Granuloma or Oral Pulse granuloma is a rare entity having unique microscopic features which is often overlooked by clinician as well as pathologist. We assume that there may have been many cases like this in the past which have got unnoticed and unreported. So we felt the urge for contributing this case in the existing literature thus enhancing the knowledge and awareness of health professionals.

Case Report: We have documented a case of 22 year old female patient with pericoronitis related with pain and swelling. On disimpaction, associated pericoronal tissue was removed and sent for histopathological assessment. Microscopically, the lesion resembled structure of processed pulses showing characteristic hyaline rings.

Conclusion: The ingested food particles impinge the wound. These food particles act as foreign matter which further induce granulomatous host response.

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1. Introduction

Pulse Granulomas (PGs) or Hyaline Ring Granulomas (HRGs) are unusual but distinct inflammatory lesions which can manifest orally and extraorally.^{1,2} Synonymously, known as giant-cell hyaline angiopathy, oral vegetable granuloma, food induced granuloma, granuloma with giant cells and hyaline change etc.¹

Histologically, lesion is typified by the presence of giant cells and pale eosinophilic structureless substance known to be hyaline rings (HR), along with areas of fibrosis with mature fibroblasts, macrophages, plasma cells and lymphocytes akin to other granulomas.^{1,2} The term "hyaline ring" was coined by Dunlap and Barker.³ Ultrastructurally, HR is visualized as a stratified structure due to the accretion of cross-banded collagen fibrils with a basement membrane-like lining containing cellulose microfilaments.⁴

Two theories have been hypothesized for etiopathogenesis:

1. Endogenous theory supports intrinsic origin, according to which, HRG arise due to hyaline degenerative changes in walls of blood vessels hence refereed as giant-cell hyaline angiopathy.³
2. Exogenous theory favours extrinsic origin and states that the hyaline rings are formed due to introduction/penetration of foreign material (pulse and legumes) into the oral mucosa or gastrointestinal tract or lungs which further provoke foreign body reaction.⁵

Knoblich R (1969) concluded that the granulomatous response is peculiarly due to cellulose moiety of the lentil particles in contrast to starch.⁶

Oral Hyaline ring granulomas have been reported to be associated with post-extraction wounds,⁷ inflammatory cyst,⁸ ameloblastoma,⁹ keratocystic odontogenic tumor¹⁰

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Here, we have documented a case of Hyaline ring Granuloma associated with pericoronitis.

2. Case Report

A 22-year-old female patient reported to our private clinic with the chief complaint of pain and swelling in right lower cheek region. Extraoral examination revealed a diffuse swelling present on the right side of the face extending 2cms away from corner of the mouth to the posterior border of the mandible and inferiorly to the lower border. On intraoral examination, pericoronitis was observed related to impacted right mandibular third molar (mesioangular) with purulent discharge which was visible from the gingival sulcus. She also presented with high fever and oedema. The patient underwent routine blood investigations followed by disimpaction and associated pericoronal tissue was sent for histopathological assessment. The patient was prescribed with antibiotics, analgesics and mouthwash.

On routine Hematoxylin and Eosin staining, the section showed aggregates of thin eosinophilic hyaline rings of varying size and shapes and were surrounded by mixed inflammatory cells along with few multinucleated giant cells [Figure 1]. The peripheral portion of the hyaline body showed positivity for Masson's trichrome stain due to condensation of collagen [Figure 2]. The patient was followed up for six months and no recurrence was noted.

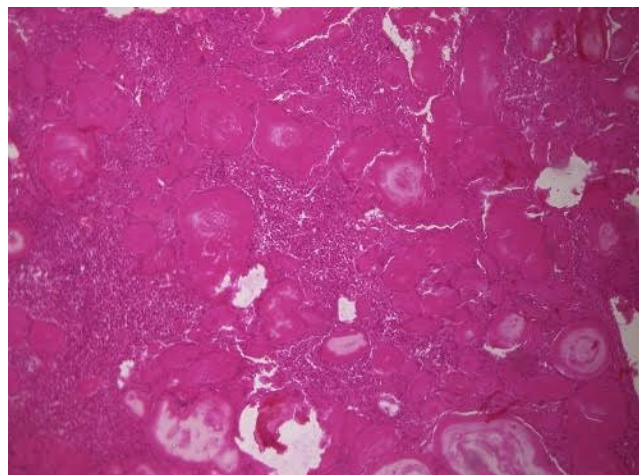


Fig. 1: The photomicrograph depicting granulomas consisting of hyaline rings. (Hematoxylin and Eosin stain, $\times 10$)

3. Discussion

PG, first introduced in 1971 by Lewars, has been a subject of much controversy regarding its nomenclature and etiopathogenesis. The term Hyaline Ring Granuloma (HRG) is the most accepted term of this lesion.¹¹ Two theories (Exogenous and Endogenous) are considered to

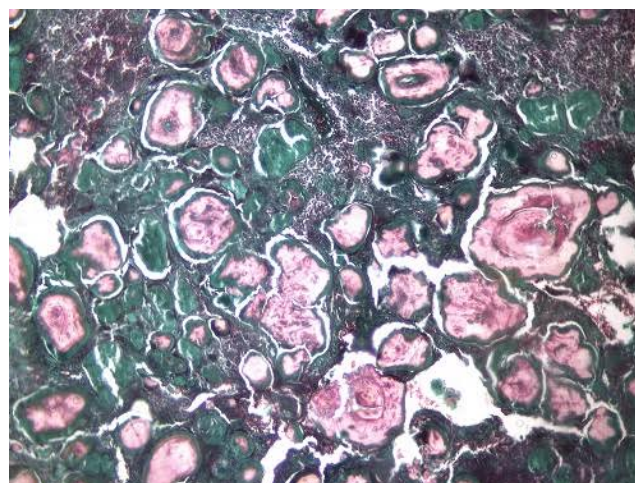


Fig. 2: The photomicrograph depicting condensation of collagen (Masson's trichrome stain, $\times 10$)

play important role in the etiopathogenesis of HRG. According to the Exogenous or Extrinsic theory, the hyaline rings are formed due to penetration and entrapment of foreign material (pulse and legumes) in the oral mucosa or gastrointestinal tract or lungs. The Endogenous or Intrinsic theory, assumes that the hyaline rings are due to the degenerative changes occurring in the wall of blood vessels particularly collagen.¹² According to Pola JG et al, the etiology is unknown although clinical and histopathologic features support inflammatory origin.⁸ The authors also reported PG in the wall of inflamed radicular cyst.⁸ In a novel study conducted by Talacko AA and Radden BG, homogenized cooked legumes were transplanted in experimental animals in the orofacial regions. They suggested that the food ingredients get altered after being digested rapidly, causing the cellulose part to be seen as hyaline rings as they are indigestible whereas starch gets digested. The host response to these structures leads to formation of granulomatous reaction.¹³ Yang et al and Chen et al, believed HRs were similar to hyaline bodies (seen in the odontogenic cysts). However, detailed morphological studies indicate that these two structures are different.^{14,15} Extraorally, the reported cases have been in lungs, wall of stomach in peptic ulcers, associated with perforated diverticulitis, buttocks, gall bladder, fallopian tube.¹ Our case, presented with a painful swelling on right posterior mandibular region. In the light of recent researches, 70% of cases have been reported as non-specific painful swelling in the posterior mandible.¹⁶ Our case showed granulomatous lesion comprising of pale eosinophilic hyaline rings along with few giant cells. This is the most characteristic feature of HRG. We analysed HRs in the lesion which were similar to the structure of the pulses. Hence we prefer the term Oral Pulse or Hyaline Ring Granuloma. Acharya S et al, mentioned that after invoking the

granulomatous response, the inflammatory enzymes modify the morphological aspects of HRs, maintaining the antigenic potential.² Histopathologically, Hyaline Ring Granuloma consists of starch granules covered with cellulose moiety along with giant cells presenting as a granulomatous lesion.¹⁰ Pola JG et al observed that immunohistochemical stains were negative for antibodies against stromal material and basement membrane hence depicting that hyaline structures are bodies of double membrane similar to walls of vegetable cells.⁸ In our case, Masson's trichome staining showed peripheral staining of the hyaline rings depicting condensation of collagen. Manjunatha BS, Kumar GS, Raghunath V observed that the hyalinized structures stained positive for PAS and van Gieson.¹⁷ In conclusion, the Oral Pulse or Hyaline Ring Granuloma is a distinct lesion which requires careful diagnosis and should be separated from other lesions.

4. Conclusion

HRGs or PGs are quite rare in occurrence and clinically they can be confused with other lesions. However, their histopathological features are quite distinct and characteristic. They can occur in conjunction with other tumors and since their presence can be overshadowed, their diagnosis can be missed. The treatment is essentially conservative and the recurrence is not reported.

5. Conflict of Interest

The authors declare no relevant conflicts of interest.

6. Source of Funding


None.

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Cite this article: Gupta A, AR. Unusual case of hyaline ring granuloma associated with pericoronitis: Case report and review of literature. *IP Indian J Anat Surg Head, Neck Brain* 2022;8(2):77-79.