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Radicular cyst with unique presentation of hyaline bodies – Case report and review of literature

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ABSTRACT

Hyaline bodies or Rushton bodies are peculiar eosinophilic, linear, curved, lamellated, glassy structures, sometimes with a granular center. They appear to be restricted to small areas of the epithelium rather than being present throughout. There is no consistent difference between different types of odontogenic cysts but are predominantly reported in radicular and dentigerous cysts. Radicular cysts are the most common inflammatory cysts found at the apices of the involved teeth and these bodies are found in approximately 10% of radicular cysts. Numerous pathogenic mechanisms regarding their origin, structure, and composition have been proposed, of which two endogenous theories gained importance, which are epithelial and hematogenous origins. Recently, β-pleated conversion of keratins as well as hemoglobin has been proposed. Majority of the hyaline bodies appear to be localized in the cystic lining, but few reports have demonstrated these bodies in the connective tissue wall, and others reported them to be straddled within basement membrane. We present a case of radicular cyst with extensive aggregation of these structures within the cystic lumen and occupying two-thirds of the thickness of cystic epithelium in a 42-year-old male patient with a review of these enigmatic structures.

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1. Introduction

Radicular cysts are the most common inflammatory cysts comprising about 52–68% of all the cysts affecting the human jaws. They arise from the epithelial residues in the periodontal ligament as a result of periapical periodontitis following death and necrosis of the pulp. Their prevalence is highest among patients in their fourth decade of life, and higher among men than women. Most of the radicular cysts are symptomless and are discovered when periapical radiographs are taken of teeth with nonvital pulps. Patient often complains of slowly enlarging swelling and is painless unless infected. Histopathology shows simple stratified squamous epithelium and arcading pattern with associated inflammatory process. Various metaplastic changes, mineralization, and hyaline bodies are found in the radicular cysts.[1] Hyaline bodies (HB's) or Rushton bodies are peculiar structures seen on surface or within the epithelial lining of odontogenic cysts. There is no consistent difference between different types of odontogenic cysts. They are predominantly present in radicular and dentigerous cysts. In approximately 10% of radicular cysts, HB's are found in epithelial lining.[1–3] They appear to be restricted to small areas of the epithelium rather than being present throughout, and cases with extensive aggregation are not reported in the literature. Hence, we would like to report a

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case of long-standing radicular cyst with dense and continuous collection of hyaline bodies within cystic epithelium and literature review of these puzzling structures.

2. Case report

A 42-year-old male patient reported to the Institution with a complaint of dull pain and swelling in the left side of upper jaw since 15 days. No significant medical problems were recorded and past dental history revealed pain in the same region 1 year back, which subsided on taking medication. The patient reported having traumatic injury several years back and noticed discoloration of left lateral incisor since 5 years. Extraoral examination revealed a solitary diffuse swelling of about 3 cm × 2.5 cm with a smooth surface. The swelling was tender and firm on palpation. The left submandibular lymph nodes were palpable and tender. On intraoral examination, there was swelling with vestibular obliteration in relation to 22, 23, 24, and 25 with buccal plate expansion. Based on clinical examination and history, a provisional diagnosis of radicular cyst was given.

Intraoral periapical radiograph of the maxillary left lateral incisor and canine region showed well-defined radiolucency and was advised for orthopantomograph (OPG). The OPG showed a large, well-corticated unilocular radiolucency involving the periapical region of 22, 23, 24 and 25 extending from the distal of the left central incisor to mesial of the left first molar (Fig. 1). Based on patient's clinical findings, and radiographic investigations, the diagnosis of radicular cyst was arrived and was subjected to enucleation with postoperative endodontic procedure. On the left side, a crevicular incision was made from the mesial surface of the left central incisor until distal surface of the first molar, the mucoperiosteal flap was raised, and the cyst was removed. The tissue specimens were sent for histopathological examination. Histopathological examination of the surgical specimen showed edematous, nonkeratinized stratified squamous epithelial lining with plenty of hyaline bodies occupying almost 2/3rds of the cystic epithelium and showing extensive aggregation of these bodies occupying entire thickness of cystic epithelium. They were of varying shapes and sizes with curvilinear, solid, circular forms and broken pieces. There were extravasated RBCs, serum exudate, hemosiderin pigment, chronic inflammatory cells, cholesterol clefts, and hyaline bodies within the lumen (Figs. 2 and 3). Few bodies showed



Fig. 1 – OPG showing unilocular radiolucency involving left maxillary region.

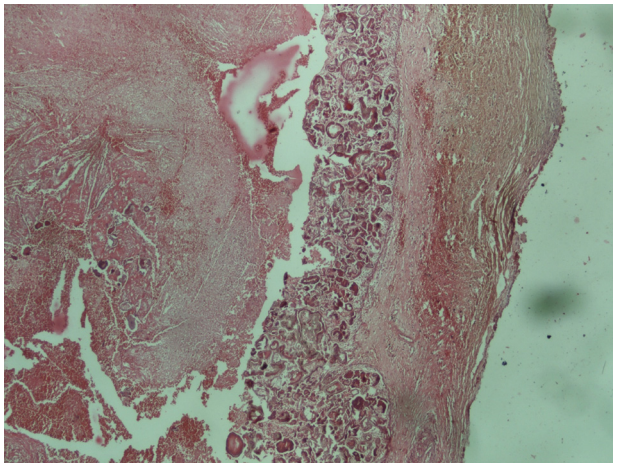


Fig. 2 – H&E section (4×) showing extensive aggregation of hyaline bodies occupying entire thickness of cystic epithelium.

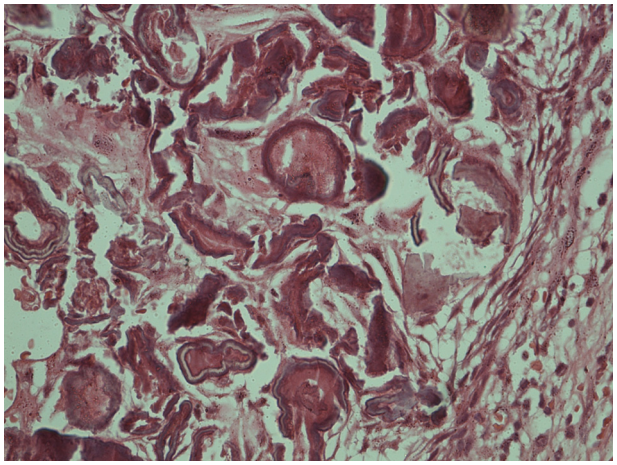


Fig. 3 – H&E section (40×) showing varying shapes and sizes of hyaline bodies with curvilinear, solid, circular forms and broken pieces.

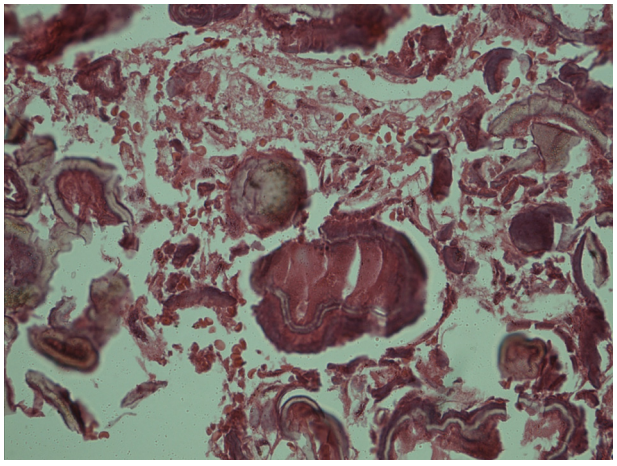


Fig. 4 – H&E section (40×) showing curvilinear bodies formed within amorphous hyaline bodies with central granular material and cell debris.

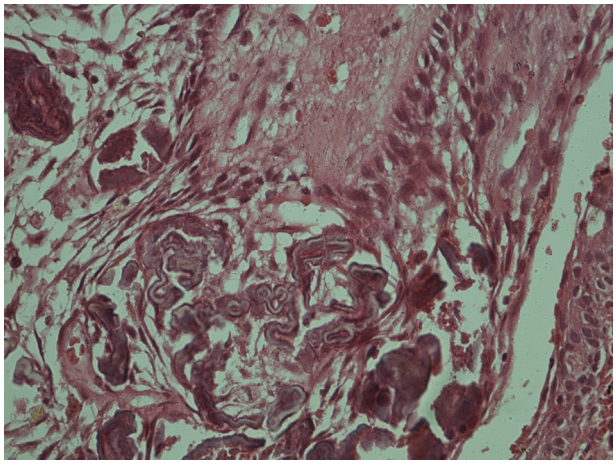


Fig. 5 – H&E section (40×) showing squamous epithelium with basal columnar, polygonal cells presenting features of spongiosis, inflammatory cells, and hyaline bodies occupying entire thickness.

curved forms within amorphous bodies (Fig. 4). The epithelium consisted of basal columnar cells and superficial polygonal to stellate cells with prominent intercellular edema and extravasated RBCs (Fig. 5). The connective tissue capsule showed collagen bundles and blood vessels with infiltration of chronic inflammatory cells. All these findings were confirmatory for the radicular cyst.

3. Discussion

HB's were first described by Dewey in 1918 and Lund in 1924.[2,4] A first detailed description of these bodies was given by Rushton in 1955 and thus they have come to be known as Rushton bodies. These bodies measure about 0.1 mm and are brittle eosinophilic or glassy structures that frequently fracture. They may be linear, straight, curved, or hairpin shaped, sometimes they are concentrically laminated. These bodies, based on their shapes, were grouped into three main types by Rushton: (a) linear, straight, or curved in double strip; (b) circular or polycyclic agglomerations often enclosing granular material; and (c) a type resembling ''broken up pieces of plate''.[5] Later Morgan and Johnson described linear, elongated needle-like bodies lining the clefts between epithelial cells and cholesterol cleft.[2]

The prevalence rate varied in different studies from 6% to 11%. They are mainly seen in radicular, dentigerous cysts and have not been described in fissural cysts.[6–9] Occasional presence of hyaline bodies in odontogenic keratocyst, lateral odontogenic cyst, calcifying odontogenic cysts, glandular odontogenic cyst, and plexiform ameloblastoma have been reported.[2,3,9–11] HB's are seen on surface or within the epithelial lining of odontogenic cysts or sometimes straddled within basement membrane. Few authors have described their occurrence in the fibrous capsule wall of dentigerous cysts, residual cysts, apical periodontal cysts, and odontogenic keratocyst.[2,3,12]

HB's commonly stained eosinophilic or remain unstained and appear glassy; rarely may they take up hematoxylin stain. Majority of the hyaline bodies appear to be localized in the epithelial lining and are present at focal areas. Their occurrence can be stated after thorough sampling of entire excisional section, and hence the prevalence varies. The epithelium around the hyaline bodies is invariably nonkeratinized stratified squamous even in odontogenic keratocyst. The thickness of the epithelium, degree of organization into layers, and rete peg pattern are variable, with some cases showing thickening. Distortion of the epithelium surrounding the hyaline bodies was also noted in some cases. Inflammatory cell infiltrate comprising predominantly of chronic inflammatory cells and few acute inflammatory cells are noted in the connective tissue wall of cysts containing HB's. Many histiocytes and multinucleated giant cells are also seen, especially in areas of extravasated RBCs and cholesterol clefts. Few studies showed no correlation between the presence of hyaline bodies and appearance of the inflammatory infiltrate.[2,3,9,13]

In contrast to the literature, the present case had continuous aggregation of hyaline bodies within cystic epithelium, which was nonkeratinized stratified epithelium, edematous with inflammatory infiltrate. The hyaline bodies were present occupying two-thirds to entire thickness of the cystic epithelium. They were of varied sizes and shapes, predominantly polycyclic forms with central granular material, and linear, curved forms. In few areas, the linear, curved forms were embedded within the polycyclic bodies suggesting they were formed from such circular agglomerations and varied morphology suggests different stages of formation. The organization of epithelium was lost due to presence of these bodies and inflammatory cells, extravasated RBCs, serum exudate, and debris were also noticed. They were not appreciated in the connective capsule in the present case. The cystic lumen contained extensive fibrinous exudate, cholesterol clefts, hyaline bodies, and foreign body giant cells. The origin, structure, and composition of these structures are debatable, and many theories have been proposed.

The histochemical studies were carried out mainly to evaluate their composition and origin, and therefore, analysis was done mainly for keratin and hemoglobin, serum proteins, fibrinogen, and fibrin. Some authors believed them to be foreign materials, such as machinery oil, fragments of pulses introduced into jaws, surgical suture materials, and cotton wool shreds.[14–16] The clear, eosinophilic nature, complete restriction to the epithelium, and no report of their presence in fissural cysts suggested them to be cuticular/keratin product, whereas others stated them to be conglutinated erythrocytes, degenerated RBC's, thrombus, serum coagula, etc. Thus two endogenous theories gained importance that they are epithelial derivatives or hematogenous.

Epithelial origin gained importance due to their resemblance of the clear material to the so-called keratinized layer of epithelial attachment of a tooth i.e., secondary enamel cuticle of Gottlieb, which in some cases are formed around degenerating cellular material. Wertheimer et al. (1962) found similar staining affinities between hyaline bodies and secondary dental cuticle.[5,7] Shear (1961) supported the theory that hyaline bodies were keratin and suggested that the granular material was probably keratohyaline, thereby implying a centripetal formation of these bodies.[6]

Dewey in 1918 put forth the concept of hyaline degeneration of capillaries leading to their formation in his original manuscript.[4] Hodson (1967), and Sedano and Gorlin (1968) suggested these bodies arose from conglutinated erythrocytes or denatured erythrocytes derived from thrombus formation in small capillaries, and so did Browne (1971) in his survey of odontogenic keratocysts.[17–19] Most of the support for hematogenous origin was derived from positive histochemical reactions with hemoglobin derivatives.[20] The enzyme histochemical studies showed lack of hydrolytic enzyme activity in adjacent epithelium, inconsistent with the concept of origin from degenerating blood vessels.[2,21]

TEM studies revealed two forms of hyaline bodies, lamellated and homogeneous. The lamellated hyaline bodies show alternate layers of electron dense and electron lucent materials with the layers ranging from two to fifteen or higher. These lamellations have been considered as appositional lines or segregation of components within the amorphous granular mass and electron dense peripheral band. The electron dense layer is usually homogenous but may occasionally be fine-granular or mottled. The electron lucent layer is granular but does reveal a meshwork structure at times. Occasional presence of small amounts of mineralization in the form of needle-like or rod-like crystals in the center has been suggested to represent dystrophic calcifications.[2,9,13,22] The lamellar pattern was considered due to segregation of various components within the mass rather than incremental deposition.[23] While others suggested that foreign material in cyst epithelium produces fine-grained matrix around coarse grain foreign material and then undergoes different degrees of homogenization.[24]

Ultrastructural findings were equally divided regarding the origin. In a recent study on hyaline bodies, Sakamoto et al. proposed that keratins and hemoglobin are released from dead epithelial cells and erythrocytes, respectively. These undergo β-sheet conversion during the process of degeneration and aggregate to form the hyaline bodies. They also put forth the concept that there is a proamyloid formation by hemoglobin before the aggregation of keratin, and that both substances were required for the generation of hyaline bodies.[25]

To conclude, the origin of hyaline bodies is obscure and their presence appears to be restricted to odontogenic cysts and tumors, indicating the role of odontogenic epithelium. The present case shows extensive aggregation of these bodies with varied morphology. The presence of these bodies does not have any clinical implication but make an interesting and unique phenomenon at cellular level.

Conflicts of interest

The authors have none to declare.

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