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Annals of Diagnostic Pathology 16 (2012) 527 – 531

Annals of DIAGNOSTIC PATHOLOGY

Crystal-storing histiocytosis: a rare lesion in periapical pathology☆,☆☆

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Abstract Crystal-storing histiocytosis is a rare manifestation of plasma cell dyscrasia/monoclonal gammopathies and lymphoproliferative disorders, characterized by cytoplasmic accumulation of crystallized immunoglobulins in histiocytes. Nevertheless, some reported cases of crystal-storing histiocytosis raise the possibility that this lesion may also be reactive. Crystal-storing histiocytosis in the oral cavity is extremely rare; only one case affecting the tongue has been reported in the English-language literature. In this report, we discuss the case of a 38-year-old man who presented a persistent periapical lesion affecting the maxillary left lateral incisor. Histopathological analysis showed numerous crystal-laden histiocytes associated with a mild plasma cell infiltrate within a fibrous stroma. The plasma cells failed to show clonal light-chain restriction, and the patient had no associated hematologic disorder or systemic disease. Thus, this lesion was probably the result of hypersecretion of immunoglobulins by polyclonal plasma cells found in the periapical lesion. Crystal-storing histiocytosis should be considered in the differential diagnosis of periapical lesions.

Keywords: Crystal-storing histiocytosis; Periapical lesion; Oral; Immunohistochemistry

1. Introduction

Crystal-storing histiocytosis (CSH) is a pathological condition characterized by accumulation of histiocytes containing paraprotein-related crystallized immunoglobulins. The disease may be systemic, usually associated with a poor prognosis, or may even be limited to a single organ. It is commonly related to underlying plasma cell dyscrasia/monoclonal gammopathies, multiple myeloma, or malignant lymphoma. Interestingly, some cases of CSH have presented as an early manifestation of multiple myeloma or malignant lymphoma at the subclinical stage. Because of this common association, an extensive clinical workup, including radiologic studies, laboratory investigations, and bone marrow biopsy, should always be performed to rule out this possibility [1-3]. In the head and neck region, several cases of CSH have been described, the majority of these being associated with either multiple myeloma or malignant lymphoma [4-8]. In the oral cavity, only a single case of CSH affecting the tongue in a patient with rheumatoid arthritis and polyclonal hypergammaglobulinemia has been reported [3]. Here, we report an extremely rare case of CSH in a periapical location.

2. Case report

The patient, a 38-year-old man, was referred to the Oral Diagnosis Center of School of Dentistry, University of

☆ Conflicts of interest: The authors deny any conflicts of interest.

☆☆ Supported by The State of São Paulo Research Foundation (FAPESP) and The National Council for Scientific and Technological Development (CNPq).

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1092-9134
http://dx.doi.org/10.1016/j.anndiagpath.2011.05.004

Ribeirão Preto, complaining of slight pain in the left anterior maxillary region with duration of 6 months. The medical history was noncontributory, and the extraoral examination showed no alterations. On intraoral examination, a purulent discharge draining in the vestibular mucosa apical to the maxillary left lateral incisor was visualized, which also showed sensitivity on palpation and to the percussion test. Radiographically, a well-circumscribed, unilocular, radiolucent lesion located in the periapical region of the maxillary left lateral incisor, measuring approximately 1.0 cm in diameter, was observed. The root canal had been endodontically treated 2 years previously, followed by a metal post restoration (Fig. 1). The maxillary left canine and first premolar responded positively to a cold pulp test. According to the clinical and radiographic features, a chronic periapical lesion, such as periapical granuloma or radicular cyst, was the main differential diagnosis. Thus, under local anesthesia, the lesion was fully excised, followed by apical surgery, without complications.

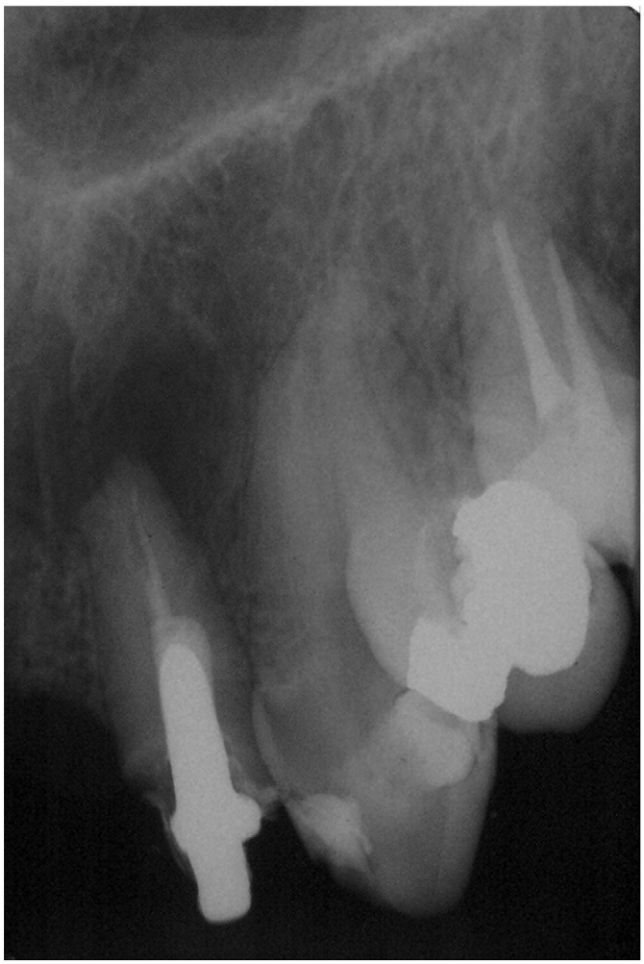

Fig. 1. Radiographic view of a well-circumscribed, unilocular, radiolucent lesion in periapical region of the maxillary left lateral incisor diagnosed as crystal-storing histiocytosis.

3. Pathologic findings

3.1. Morphologic findings and histochemical analysis

The specimen measuring 1.0 × 0.9 cm was fixed in 10% formalin and routinely processed for histological examination. Histological sections stained with hematoxylin and eosin (H&E) showed a nodular lesion containing numerous large epithelioid eosinophilic cells within a fibrous stroma intermingled with a mild plasmacytic infiltrate and scattered lymphocytes distributed around slitlike vascular structures (Fig. 2A, B). At higher magnification, the bright eosinophilic cytoplasm of the epithelioid-shaped cells seemed to be filled with numerous needlelike crystals and exhibited focal areas of birefringence under polarized light microscopy (Fig. 2C, D). The nuclei were small and bland, and there were no mitoses. Other sections stained for periodic acid–Schiff, with and without diastase digestion, were negative. Moreover, stains for acid-fast bacilli and fungi failed to reveal infectious microorganisms.

3.2. Immunohistochemistry and in situ hybridization

Tissue sections 3-μm thick were placed on silanized slides and were stained using the avidin-biotin peroxidase complex technique and heat-induced epitope retrieval buffer. The antibodies used included CD3, CD20, CD34, CD43, CD45, CD68 (PG-M1 and KP1), CD79a, CD117, CD138, myeloperoxidase, epithelial membrane antigen, plasma cell marker (VS38c), S100, heavy chain for immunoglobulin (Ig) G, and κ and λ light-chain immunoglobulins (DAKO, Carpinteria, CA). The crystal-laden epithelioid-shaped cells showed positivity for both CD68 (KP1) (Fig. 3A) and CD68 (PG-M1), confirming their histiocytic origin. CD68 (KP1) immunostained stronger than CD68 (PG-M1). The surrounding typical plasma cells were positive for CD138 (Fig. 3B), CD79a, epithelial membrane antigen, VS38c, IgG (Fig. 3C), and κ and λ light-chain immunoglobulins (Fig. 3D, E). The intracytoplasmic crystal-like inclusions showed weak immunoreactivity for IgG and κ and λ light-chain immunoglobulins (Fig. 3C-3E). CD45, CD3, and CD43 showed scarce T-cell lymphocytes, whereas CD20 was negative. The slitlike vessels were evidenced with CD34, whereas CD117, myeloperoxidase, and S100 showed scarce mast cells, neutrophils, and dendriticlike cells, respectively. In situ hybridization for Epstein-Barr virus–encoded small nuclear RNA (EBER) complementary to Epstein-Barr virus EBER1 and EBER2 loci (EBER, PNA probes, DAKO, Glostrup, Denmark) was negative.

3.3. Scanning electron microscopy

Scanning electron microscopy (Jeol JSM 5600 LV, Dental School of Piracicaba-UNICAMP, Piracicaba, Brazil) using a 5-μm paraffin section for tissue morphology evaluation revealed numerous histiocytes showing several cytoplasmic prolongations blending with each other.