Case Report

Langerhans cell histiocytosis simulating endodontic periapical lesion: Are we prepared to diagnose and manage it? A case report

ABSTRACT

Many aggressive non-endodontic radiolucent lesions show very similar clinical and radiographical features to periapical lesions of endodontic origin. Since the treatments of endodontic and non-endodontic lesions differ markedly, a precise diagnosis is imperative. Thus, the present study aimed at presenting a clinical case on the diagnosis and management of a Langerhans cell histiocytosis (LCH) lesion mimicking a periapical lesion of endodontic origin. A 51-year-old male patient was referred to a private dental office due to slight pain from the region of tooth 36. Although no sign of prosthetic or endodontic failure was noted, radiographical examination revealed a radiolucent image with poorly defined borders associated with the periapical region of the tooth. Apicoectomy and bone curettage were then performed and, given the clinical and laboratory features, the definitive diagnosis of solitary eosinophilic granuloma was made. The surgical treatment was sufficient for the remission of the symptoms, and recurrence was not observed. Given the current case, dentists should be aware of LCH lesions as they may mimic endodontic periapical pathoses, leading to misdiagnosis and therapeutic complications. Moreover, alveolar bone lesions may be the first or only sign of LCH in many cases.

Keywords: Eosinophilic granuloma, Langerhans cell histiocytosis, mandible, periapical diseases

INTRODUCTION

Periapical lesions of endodontic origin are the most common pathologic conditions affecting alveolar bone.[1] Radiolucent lesions resulting from dental pulp necrosis may be histopathologically classified into radicular cysts, periapical granulomas, and periapical abscesses[2]; however, other non-endodontic affections with somewhat similar features such as keratocystic odontogenic tumors, central giant cell lesions, ameloblastomas, and metastatic lesions are less frequently encountered.[3]

A precise diagnosis of periapical pathoses is imperative since the treatments of endodontic and non-endodontic lesions differ markedly.[2] This process, however, is not simple many times, especially for cases in which non-endodontic lesions are located in the periapical region of teeth presenting pulp necrosis.[1] Misdiagnosis of pathologies mimicking endodontic periapical lesions may even lead to considerable morbidity and even mortality.[4]

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As periapical lesions are routinely managed in dentistry, few studies have addressed their peculiarities, including detailed information about the frequency of those not associated with pulpal necrosis.[4] Moreover, regardless of the geographic differences, data from diagnostic biopsy services seems to be biased in several respects, i.e., the number and nature of

Marcelo Marcucci, Rafael Verardi Serrano[1], Luana Campos[2], Luiz Felipe Palma[1]

Stomatology and Oral and Maxilofacial Surgery Center, Hospital Heliópolis, [1]Graduate Dentistry Program, Ibirapuera University, [2]Graduate Program in Implantology, University of Santo Amaro, School of Dentistry, São Paulo, SP, Brazil

Address for correspondence: Prof. Luiz Felipe Palma, Ibirapuera University, Av. Interlagos, 1329 - Chácara Flora, São Paulo, SP, 04661-100, Brazil.
E-mail: luizfelipep@hotmail.com

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the specimens to be evaluated may vary according to the clinical practice of the contributors.[5]

Langerhans cell histiocytosis (LCH) is a clonal neoplastic proliferation of Langerhans type cells, dendritic cells present in skin and mucosa,[6] resulting in tissue destruction secondary to cellular infiltration.[7,8] The concomitant presence of a varying number of leucocytes, eosinophils, neutrophils, lymphocytes, plasma cells, and giant multi-nucleated cells is also seen.[7] Also historically named histiocytosis X, the pathogenesis of LCH remains uncertain.[9]

LCH comprises chronic focal LCH or eosinophilic granuloma (bone lesions without visceral involvement), chronic diffuse LCH or Hand-Shuller-Christian disease (bone, skin, and viscera involvements), acute disseminated LCH or Letterer-Siwe disease (rapidly progressing pathology of aggressive behavior, with skin, viscera, and bone marrow involvements),[7,10] and congenital reticulohistiocytosis (only skin and mucosa involvements).[7] Eosinophilic granuloma is the most prevalent form of LCH (60 to 70%) and is restricted to bones, manifesting as solitary or multifocal bone lesions.[9]

In light of these facts, the present study aims at presenting a clinical case on the diagnosis and management of an LCH lesion mimicking a periapical lesion of endodontic origin.

CASE REPORT

A 51-year-old male patient was referred to a private dental office due to slight pain from the region of tooth 36. It had been treated endodontically many years ago and received a metal-ceramic crown with metal cast posts and core. Although no sign of prosthetic or endodontic failure was noted, radiographical examination revealed a radiolucent image with poor-defined borders associated with the periapical region of the tooth [Figure 1]. Considering the diagnostic hypothesis of an infectious/inflammatory lesion of endodontic origin, surgical therapy with apicectomy (both roots) and bone curettage was proposed.

The bone material collected was sent for histopathological analysis and showed histiocytic proliferation with scattered and intermingled multinucleated eosinophilic giant cells [Figure 2a and b]. Furthermore, immunohistochemically, there was positivity to S100 [Figure 3a and b] and lysozyme.

The patient was submitted to a systemic medical investigation but no further involvement was found. Thus, given the clinical and laboratory features, the definitive diagnosis of Langerhans cell histiocytosis, presented as a solitary eosinophilic granuloma, was made.

After 10 years and without any symptoms, the patient returned to the office. A limited acute abscess with fistula was noted clinically and, radiographically, a new radiolucent image associated with the periapical region of the same tooth was seen [Figure 4]. The tooth was then extracted, the lesion was curetted, and the bone material collected was sent for histopathological analysis again. Microscopically, a nonspecific chronic inflammatory process was observed with areas of scarring fibrosis. Following this surgical treatment, no further complaint was present anymore.

Within 2 years of follow-up, the patient received a dental implant for rehabilitation, with success.

DISCUSSION

The current study presents the diagnosis and management of an LCH single bone lesion mimicking a periapical lesion of endodontic origin. It can be considered interesting because the periapical region affected was related to a

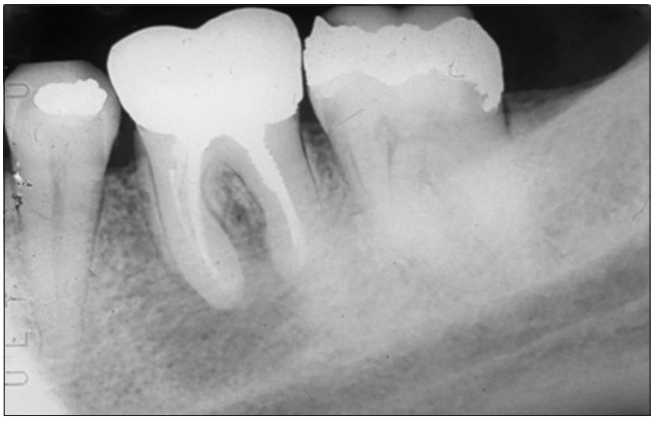

Figure 1: Periapical radiography: radiolucent image with poor-defined borders associated with the periapical region of the tooth 36

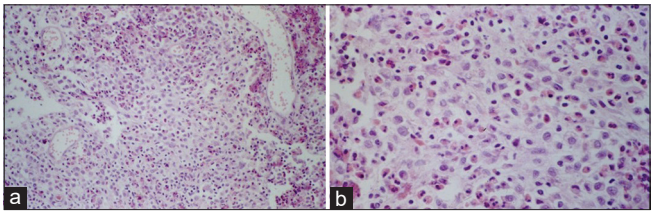

Figure 2: Histopathological analysis: histiocytic proliferation with scattered and intermingled multinucleated eosinophilic giant cells. (a) 40x, (b) 100x

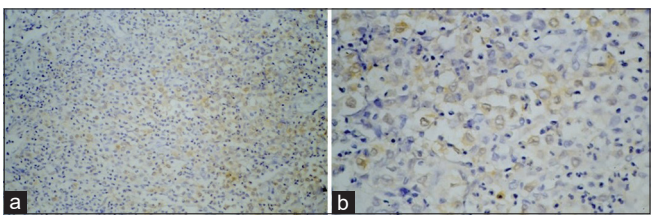

Figure 3: Immunohistochemical analysis: positivity to S100. (a) 40x, (b) 100x

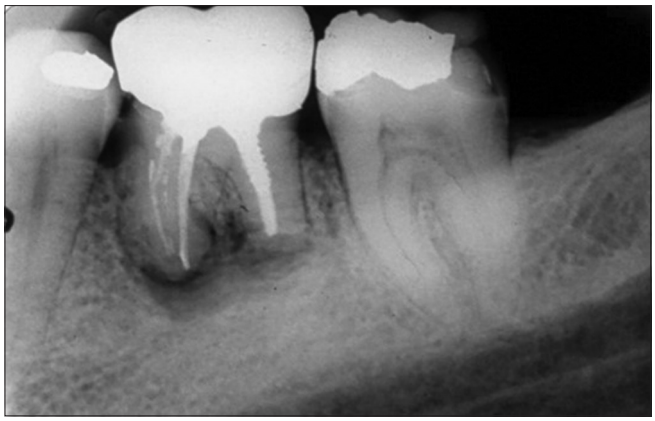

Figure 4: Periapical radiography: radiolucent image associated with the periapical region of the tooth 36

tooth endodontically treated many years ago, leading to a challenging diagnosis. To the best of the authors' knowledge, there are too few similar reports in the literature.

Eosinophilic granuloma rarely affects adults; however, when encountered, it is more common from the first to the third decades of life[9] in men.[11] The jaws are involved in 10 to 20% of the cases, with a higher predilection for the mandible,[6] and the diagnosis should be made according to clinical, radiographical, and histological examinations.[7]

Clinical presentation and oral symptoms vary among the age groups, including pain, swelling, loosening of teeth, and limitation of mouth opening. No complaint may also be reported. Radiographically, the lesions show variable, not specific radiographic features but, generally, they are radiolucent and present well-defined borders.[9] Perforation of the cortical plate may be seen in larger lesions.[8]

Conventional microscopy reveals conjunctive fibrous tissue areas associated with a mixed inflammatory infiltrate, as well as non-malignant histiocytic proliferation along with the Langerhans cells.[7] By immunohistochemical techniques, lesional Langerhans cells present positive to CD1A, langerin, S100, CD68, vimentin, HLA-DR, CD45, CD4, and lysozyme.[6] No specific laboratory analysis or test exists for LCH diagnosis but early detection and searching for other tissues/organ involvement is critical.[7]

The prognosis of LCH depends on the stage of the disease (survival rates of about 33% in cases of multiorgan involvement),[6] but it is generally good for cases of a single LCH bone lesion.[10] The treatment of eosinophilic granuloma is performed surgically by enucleation and curettage or using radiotherapy, chemotherapy, or intralesional corticosteroid injections. Spontaneous regression is not frequent.[9]

The management herein used was based on the main diagnostic hypothesis of a periapical lesion due to failure of endodontic treatment. Fortunately, surgical apicoectomy and bone curettage were sufficient for the remission of the symptoms, and recurrence was not observed within a 10-year follow-up period. The patient did also not show any other lesion or organ involvement and, despite tooth extraction after some years due to endodontic failure, an implant rehabilitation could be performed without complications.

CONCLUSION

Given the case presented, dentists should be aware of LCH lesions as they may mimic endodontic periapical pathoses, leading to misdiagnosis and therapeutic complications. Moreover, alveolar bone lesions may be the first or only sign of LCH in many cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his 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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