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. Radiolucent lesions resulting from dental pulp necrosis may be histopathologically classified into radicular cysts, periapical granulomas, and periapical abscesses; 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.

A precise diagnosis of periapical pathoses is imperative since the treatments of endodontic and non-endodontic lesions differ markedly. 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. Misdiagnosis of pathologies mimicking endodontic periapical lesions may even lead to considerable morbidity and even mortality.

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

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 narrow involvements), and congenital reticulohistiocytosis (only skin and mucosa involvements). Eosinophilic granuloma is the most prevalent form of LCH (60 to 70%) and is restricted to bones, manifesting as solitary or multifocal bone lesions.

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 curedtted, 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...
Marcucci, et al.: LCH mimicking a periapical lesion

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.

Financial support and sponsorship
Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Huang H-Y, Chen Y-K, Ko EC-C, Chuang F-H, Chen P-H, Chen C-Y, et al. Retrospective analysis of nonendodontic periapical lesions misdiagnosed as endodontic apical periodontitis lesions in a population of Taiwanese patients. Clin Oral Investig 2017;21:2077-82.
2. Kosanwat T, Poomsawat S, Kitisubkanchana J. Non-endodontic periapical lesions clinically diagnosed as endodontic periapical lesions: A retrospective study over 15 years. J Clin Exp Dent 2021;13:e586‑93.
3. Koivisto T, Bowles WR, Rohrer M. Frequency and distribution of radiolucent jaw lesions: A retrospective analysis of 9,723 cases. J Endod 2012;38:729‑32.
4. Sullivan M, Gallagher G, Noonan V. The root of the problem. J Am Dent Assoc 2016;147:646‑9.
5. Daley TD, Wysocki GP, Pringle GA. Relative incidence of odontogenic tumors and oral and jaw cysts in a Canadian population. Oral Surg Oral Med Oral Pathol 1994;77:276‑80.
6. Peters SM, Pastagia J, Yoon AJ, Philippone EM. Langerhans cell histiocytosis mimicking periapical pathology in a 39-year-old man. J Endod 2017;43:1909‑14.
7. Madrigal-Martínez-Pereda C, Guerrero-Rodríguez V, Guisado-Moya B, Meniz-García C. Langerhans cell histiocytosis: Literature review and
descriptive analysis of oral manifestations. Med Oral Patol Oral Cir Bucal 2009;14:E222-8.
8. Lin LM, Wyman TP, Bushell A, Langeland K. Eosinophilic granuloma of the jawbone. J Endod 1979;5:25-30.
9. Lee B-D, Lee W, Lee J, Son H-J. Eosinophilic granuloma in the anterior mandible mimicking radicular cyst. Imaging Sci Dent 2013;43:117-22.
10. Kuo YS, Wu YH, Sun A, Chiang CP. Eosinophilic granuloma of the mandible mimicking a periapical lesion. J Dent Sci 2017;12:424-5.
11. Pringle GA, Daley TD, Veinot LA, Wysocki GP. Langerhans’ cell histiocytosis in association with periapical granulomas and cysts. Oral Surg Oral Med Oral Pathol 1992;74:186-92.