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Mandibular fracture due to rare manifestation of Langerhans cell histiocytosis

Fratura patológica da mandíbula por rara manifestação de histiocitose de células de Langerhans

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Dear Editor,

Langerhans cell histiocytosis (LCH) is reported as being rare in the literature, with an incidence of 5 cases per 1 million per year and a prevalence of 3.7:1 in men:women. LCH is generally limited to an organ, most commonly lesions of the bone, whether solitary or multiple, that tend to appear in the cranial or femoral region in children younger than 10 years and in the costal arches, scapula or mandible in patients up to 20 years of age.[1-3] The diagnosis is based on clinical symptoms, radiographic symptoms and especially histological examination after biopsy.

We present a rare manifestation in a male patient, 51 years old, with clinical and radiographic examinations presenting a pathological fracture of the mandible on the right side (Figure 1), due to a radiolucent lesion of approximately 2.5cm, that was circumscribed and well delimited, and the presence of another lesion in the symphysis region that was approximately 0.5cm in diameter. The histological finding was consistent with the infiltration of Langerhans cells, macrophages, lymphocytes, eosinophils, granulocytes and giant cells, that positively reacted to immunohistochemistry for CD-1a, CD207 (Langerin) and Protein S100 (Figure 2A - C).[4,5]

The rapid evolution and aggressiveness of the lesion, as well as the initial diagnostic hypothesis of being a malignant lesion, indicated that treatment should involve hospital admission and multidisciplinary planning, which increases the time of treatment and costs because the patient will need reconstructive surgery to restore his functional and aesthetic quality of life.

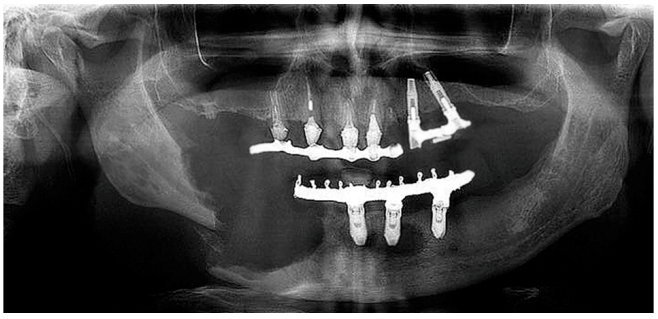

Figure 1 - Panoramic radiography presenting a pathological fracture of the mandible on the right side.

Conflicts of interest: None.

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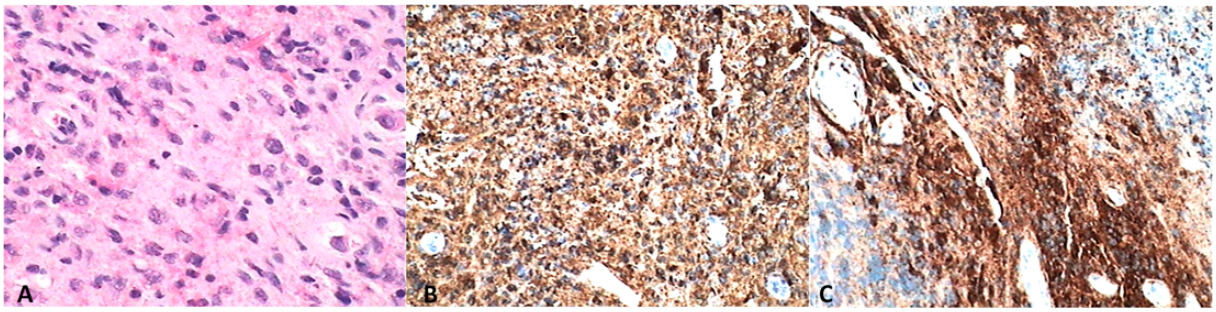

Figure 2 - Histological exam. (A) Eosin and hematoxylin; (B-C) Positive reaction to immunohistochemistry by CD-1a, CD207 (Langerin) and Protein S100.

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REFERENCES

1. Rees J, Paterson AW. Langerhans cell histiocytosis in an adult. Br J Oral Maxillofac Surg. 2009;47(1):52-3.
2. Nicollas R, Rome A, Belaïch H, Roman S, Volk M, Gentet JC, et al. Head and neck manifestation and prognosis of Langerhans' cell histiocytosis in children. Int J Pediatr Otorhinolaryngol. 2010;74(6):669-73.
3. Peters SM, Pastagia J, Yoon AJ, Philipone EM. Langerhans cell histiocytosis mimicking periapical pathology in a 39-year-old man. J Endod. 2017;43(11):1909-14.
4. Merglová V, Hrušák D, Boudová L, Mukenšnabl P, Valentová E, Hostička L. Langerhans cell histiocytosis in childhood - review, symptoms in the oral cavity, differential diagnosis and report of two cases. J Craniomaxillofac Surg. 2014;42(2):93-100.
5. Laird J, Ma J, Chau K, Chelius M, Shi W, Zhang Z, et al. Outcome after radiation therapy for Langerhans cell histiocytosis is dependent on site of involvement. Int J Radiat Oncol Biol Phys. 2018;100(3):670-8.