

Intraosseous Myofibroma of the Mandible: A Case Report

Miofibroma Intraoseo Mandibular: Reporte de Caso

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ABSTRACT: The myofibroma is defined as a benign neoplasm formed by myoid contractile cells localized around the wall of thin blood vessels, it is a tumor that presents itself mostly in infancy although it may occur at any age and it is most common in head and neck; it is uncommon to be localized in the oral cavity and even less common if it is intraosseous. It may also be associated to miofibromatosis or present itself as a solitary lesion. The differential diagnosis depends on the localization and the radiographic characteristics; it would be very difficult to include, at first hand, myofibroma as an intraosseous lesion. Histopathologically, a neoplasm with a biphasic pattern formed by spindle cells in fascicles and bundles, spindle nucleus with eosinophilic cytoplasm inside a hyalinized stroma was found. In lesions of neoplasms of spindle cells histological studies should be supported by an immunohistochemical panel and show positive results to antibodies Actin, smooth muscle Actin and Vimentin.

KEY WORDS: myofibroma, intraosseous, mandible.

INTRODUCTION

The myofibroma is defined as a benign neoplasm formed by myoid contractile cells localized around the wall of thin blood vessels (Fletcher *et al.*, 2002), it is a tumor that presents itself mostly in infancy although it may occur at any age and it is most common in head and neck; it is uncommon to be localized in the oral cavity and even less common if it is intraosseous (Poon & Kwan, 2005; Kin *et al.*, 2006; Acosta *et al.*, 2012; Sundaravel *et al.*, 2013).

CASE REPORT

A forty-five-year-old female shows up for dental caries treatment of chronic evolution with light pain. The intraoral examination shows an extensive second

degree carious process on dental organ (DO) 37, with a light growth of the vestibular cortical compared to the contralateral side and soft tissue similar coloration to the adjacent mucous; an orthopantomography and periapical radiography were requested. A well-defined radiolucid lesion with sclerotic surrounding and affection to the distal root of DO 37 was observed; a surgical extraction was decided (Fig. 1). Based on the carious process and the radiographic findings, a diagnosis of periapical granuloma vs. periapical cyst was considered. The excisional biopsy was performed.

In the histopathological study, a neoformation of non-encapsulated, well vascularized spindle cells of undulated nucleus, arranged in a "lace-like pattern" over a stroma of dense connective tissue and a mixed inflammatory infiltrate was observed (Fig. 2, a and b). Was positive to antibodies muscle actin specific (clone

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HHF35) 1:50, smooth-muscle actin (clone 1A4), CD34 (clone QBend34) positive in the peripheral vessels, but negative in the neoplasm 1:50 and positive for vimentin (clone V9) 1:100 (Fig. 3 a, b, c, d) and negative to antibodies s100 (polyclonal) 1:100 and desmin (clone D33) 1:100 in the immunohistochemical panel. An intraosseous myofibroma was concluded.



Fig. 1. Ortopantomography requested from the patient for the clinical-radiographic evaluation. Radiolucid area localized at level of DO 37.

Fig. 2. Histopathological Features. H-E a) 200x Lesion of mesenchymal lineage, not encapsulated, very cellular, formed by fibro-connective tissue with dense irregular arrangement of bundles of collagen fibers. b) 400x spindle cells of undulated nucleus, arranged in a "lace-like pattern" and a mixed inflammatory infiltrate.

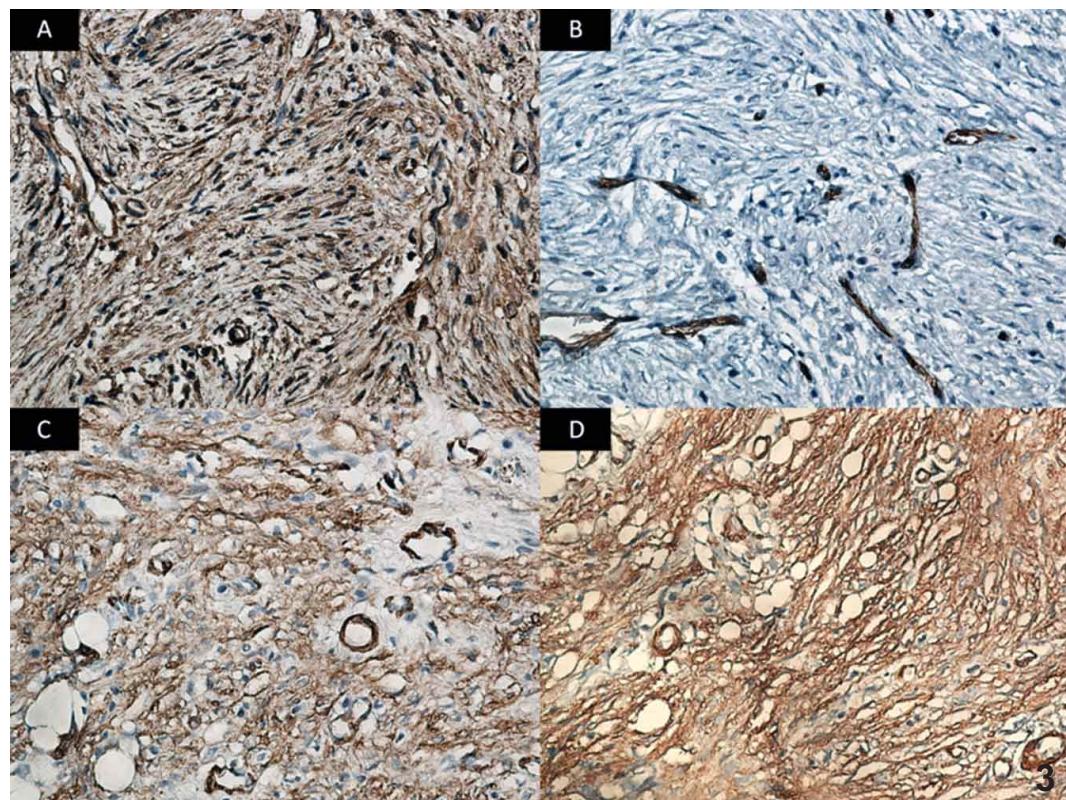
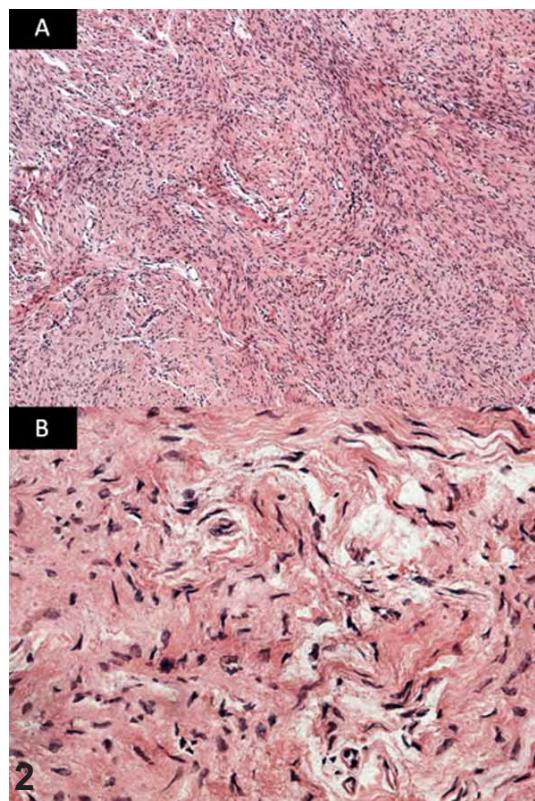


Fig. 3.
Immunohistochemical panel. 400x a) Mesenchymal tissue intensely positive to Vimentin, b) Antibody CD 34 negative in the neoplasm and positive in the peripheral vessels, c y d) Identification of myofibroblasts through antibodies muscle actin specific and smooth muscle actin.

DISCUSSION

The etiology of myofibroma is uncertain; it may be of autosomal dominant or recessive type; nevertheless, the family incidence is low. It is suggested that there might be other factors involved (Kin *et al.*; Souza *et al.*, 2009; Acosta *et al.*). It may also be associated to myofibromatosis or present itself as a solitary lesion as in the case presented. It is a tumor that affects the head and neck more frequently, although it is not so common in the oral cavity and less common intraosseously in the mandible (Acosta *et al.*) as in this report. In the vast series of cases reported by Foss & Ellis (2000), 79 myofibromas in the oral region, the mandible was implicated in a third; 12 were central lesions and the cortical or the surface of the periosteum in 15 were affected. Brasileiro *et al.* (2010) mentions the development of this neoplasm at early ages. Allon *et al.* (2007) observed that from 19 cases of mandible intraosseous myofibroma in the literature and four more included at the time, the mean age was 7.2 years; this paper agrees with other authors that have reported solitary mandible myofibroma in adult ages in the third and fourth decades of life (Oliver *et al.*, 2003; Ramadorai *et al.*, 2010; Sundaravel *et al.*; Brierley *et al.*, 2012).

The differential diagnosis depends on the localization and the radiographic characteristics (Acosta *et al.*), it would be very difficult to include, at

first hand, myofibroma as an intraosseous lesion and it would have to be thought as a cyst, as it is this case, even if the patient referred pain due to caries. The myofibroma was a radiographic finding. Histopathologically, a neoplasm with a biphasic pattern formed by spindle cells in fascicles and bundles, spindle nucleus with eosinophilic cytoplasm inside a hyalinized stroma was found (Poon & Kwan; Kin *et al.*; Brasileiro *et al.*; Nirikalpa & Narayanan, 2011; Acosta *et al.*). The myofibroma is positive to antibodies Actin, smooth muscle Actin and Vimentin; it is negative to Desmin S100 and EMA (Shields *et al.*, 1998; Shibuya *et al.*, 2008; Souza *et al.*; Brasileiro *et al.*; Nirikalpa & Narayanan; Acosta *et al.*).

Surgical treatment is required and although the neoplasm is not encapsulated, the relapse is rare (Poon & Kwan; Kin *et al.*; Sundaravel *et al.*; Cargini *et al.*, 2012; Abramowicz *et al.*, 2012), in the case presented, there has been no relapse after a year.

CONCLUSIONS

In lesions of neoplasms of spindle cells histological studies should be supported by an immunohistochemical panel.

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RESUMEN: El miofibroma es una neoplasia benigna compuesta por células mioides contráctiles localizadas alrededor de la pared de vasos sanguíneos delgados, es un tumor que se presenta sobre todo en la infancia aunque puede ocurrir a cualquier edad, tiene predilección en cabeza y cuello, sin embargo en cavidad oral es raro y aún más si es intraóseo, puede estar asociado a miofibromatosis o bien presentarse de manera solitaria. Los diagnósticos diferenciales dependen de la localización y de las características radiográficas y de primera instancia es muy difícil incluir al miofibroma entre las lesiones intraóseas. Histológicamente presenta patrón bifásico conformado por células fusiformes dispuestas en fascículos y haces así como núcleos fusiformes con citoplasma eosinófilo dentro de un estroma hialinizado. Es necesario recurrir al panel de inmunohistoquímica en neoplasias de células fusiformes, positivo a Acs Actina, Actina músculo liso y Vimentina. Reportamos el caso de una mujer de 45 año con un miofibroma en la mandíbula.

PALABRAS CLAVE: miofibroma, intraóseo, mandíbula.

REFERENCIAS BIBLIOGRÁFICAS

Abramowicz, S.; Simon, L. E.; Kozakewich, H. P.; Perez-Atayde, A. R.; Kaban, L. B. & Padwa, B. L. Myofibromas

of the jaws in children. *J. Oral Maxillofac. Surg.*, 70(8):1880-4, 2012.

- Acosta, R. M.; Castro, G. F.; Monroy, H. V.; González, B. J. & López, S. F. Miofibroma intraóseo mandibular en niños. *Rev. Mex. Cir. Bucal Max.*, 8(2):51-5, 2012.
- Allon, I.; Vered, M.; Buchner, A. & Dayan, D. Central (intraosseous) myofibroma of the mandible: clinical, radiologic, and histopathologic features of a rare lesion. *Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod.*, 103(4):e45-53, 2007.
- Brasileiro, B. F.; Martins-Filho, P. R.; Piva, M. R.; da Silva, L. C.; Nonaka, C. F. & Miguel, M. C. Myofibroma of the oral cavity. A rare spindle cell neoplasm. *Med. Oral Patol. Oral Cir. Bucal*, 15(4):e596-600, 2010.
- Brierley, D. J.; Khurram, S. A. & Speight, P. M. Solitary myofibroma of the adult mandible: a case report. *Oral Surg. Oral Med. Oral Pathol. Oral Radiol.*, 115(3):e40-3, 2013.
- Cargini, P.; Fidanza, F.; Facente, M. V.; Sgolastra, F.; Gatto, R. & Cutilli, T. Gingival myofibroma. A case report. *Eur. J. Paediatr. Dent.*, 13(1):81-3, 2012.
- Fletcher, C. D. M.; Unni, K. K. & Mertens, F. *Pathology and Genetics of Tumours of Soft Tissue and Bone*. World Health Organization Classification of Tumours. Lyon, IARC Press, 2002. pp.59-61.
- Foss, R. D. & Ellis, G. L. Myofibromas and myofibromatosis of the oral region: a clinicopathologic analysis of 79 cases. *Oral Surg. Oral. Med. Oral Pathol. Oral Radiol. Endod.*, 89(1):57-65, 2000.
- Kin, J. S.; Kim, S. E. & Kim, J. D. Myofibroma of the mandible: A case Report. *Korean J. Oral Maxillofac. Radiol.*, 36:211-5, 2006.
- Nirvikalpa, N. & Narayanan, V. Intraosseous infantile myofibroma of the mandible. *Ann. Maxillofac. Surg.*, 1(1):87-90, 2011.
- Oliver, R. J.; Coulthard, P.; Carre, C. & Sloan, P. Solitary adult myofibroma of the mandible simulating an odontogenic cyst. *Oral Oncol.*, 39(6):626-9, 2003.
- Poon, C. & Kwan, P. Myofibroma of the Mandible: A case Report. *Chin. J. Oral Maxillofac. Surg.*, 16:156-65, 2005.
- Ramadorai, A.; Rajsekaran, A. & Narayanan, V. A Case Report of solitary, intraosseous, Adult-Onset Myofibroma of the mandible. *J. Maxillofac. Oral Surg.*, 9(3):280-3, 2010.
- Shibuya, Y.; Takeuchi, J.; Sakaguchi, H.; Yokoo, S.; Umeda, M. & Komori, T. Myofibroma of the Mandible. *Kobe J. Med. Sci.*, 54(3):169-72, 2008.
- Shields, C. L.; Husson, M.; Shields, J. A.; Mercado, G. & Eagle, R. C. Jr. Solitary intraosseous infantile myofibroma of the orbital roof. *Arch. Ophthalmol.*, 116(11):1528-30, 1998.
- Souza, D. P.; Loureiro, C. C.; Rejas, R. A.; Souza, S. O. & Raitz, R. Intraosseous myofibroma simulating an odontogenic lesion. *J. Oral Sci.*, 51(2):307-11, 2009.
- Sundaravel, S.; Anuthama, K.; Prasad, H.; Sherlin, H. J. & Ilavaraja, V. Intraosseous myofibroma of mandible: A rarity of jaws: With clinical, radiological, histopathological and immunohistochemical features. *J. Oral Maxillofac. Pathol.*, 17(1):121-5, 2013.

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