Mandibular intraosseous leiomyoma in a child: report of a case

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Leiomyoma is a benign tumor of smooth muscle origin that rarely occurs in the oral cavity. Of the 118 cases occurring in the oral cavity described since 1884, only five involved the mandible. This is a case of an intraosseous leiomyoma in a 9-year old child, who presented to our attention with a firm intra-oral mass, involving the angle and the posterior portion of the left mandibular body. The patient was treated with local incision alone, preserving the permanent dental germs and the left inferior alveolar nerve. He remained disease free with regular follow-up for 40 months.
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INTRODUCTION

Leiomyoma is a benign tumor of smooth muscle origin that rarely occurs in the oral cavity. It usually arises in the female genital tract (95% of cases), followed by gastrointestinal tract and subcutaneous tissues.[1]

In 1975, Farman found only five tumors located in the oral cavity.[2] The first oral case was reported by Blanc in 1884[5] and two extensive reviews by Natiella *et al*.[4] and Praal *et al*.[5] from 1884 to 1982 revealed a total of 78 different cases of oral leiomyoma. In addition, a review of the Japanese literature[6] from 1957 to 1982 revealed 28 cases not included in the previous review, together with two cases of angioleiomyoma of the mandible described by White *et al* in 1985.[7]

The tongue, the lips, the hard and soft palate are the most commonly involved sites, the floor of the mouth, the major salivary glands, the alveolar maxillary bone, and the mandible are rarely involved. Of the 118 cases occurring in the oral cavity described since 1884, only 5 involved the mandible.[1,2]

This present report describes an intraosseous leiomyoma arisen within the mandible in a 9-year-old child.

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CASE REPORT

In September 1999, a 9-year-old boy was referred to our Department with a painless swelling of the left hemimandible, noticed by his family dentist two months previously. The patient had first noted increasing mobility of the left 2nd lower molar and a small lobulated mass in the region. There was no history of trauma to the area.

Intraoral examination revealed a firm, elastic mass with a diameter of 2cm involving the angle and the posterior portion of the left mandibular body. The overlying mucosa was intact and had a normal appearance. Regional lymph nodes were not involved. General examination did not reveal any other abnormalities.

The panoramic radiograph did not display any radiolucency. Only die CT scan examination showed, a solid, round tumor, which appeared to expand the external mandibular coxtex from the 2nd deciduous molar to the 1st permanent molar. The unilocular, lucency was approximately 1.8cm x 1.3 cm. There was no resorption of the left molar roots (Figure 1).

In the light of the clinical and radiographic report and for the young age of thc patient, it was decided that a limited local excision of the lesion should be the treatment of choice.

Under general anesthesia, the lesion was exposed via an intraoral approach and excised in continuity with a small margin of normal bone. The mass was dissected far from the left inferior alveolar nerve, which was found to pass laterally out of the lesion. The permanent dental germs were preserved and the wound was closed primarily.

On histological examination, the resected tumor was diagnosed as an intraosseous leiomyoma. The tissue specimens were fixed in 10% neutral formalin, and were routinely processed for light microscopy. Sections were stained with hematoxylin and eosin (Figure 2).

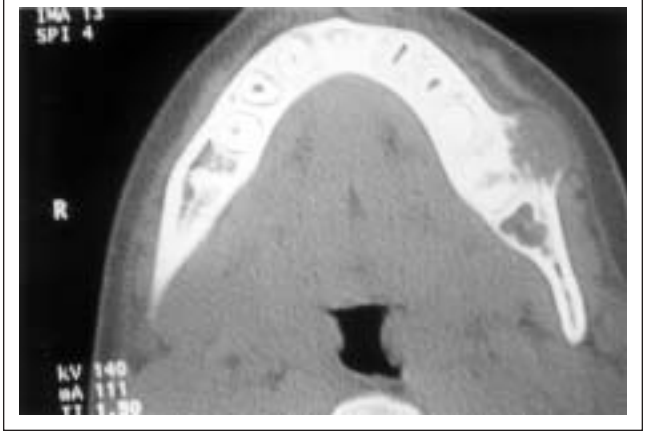

Figure 1. Axial CT displays the round mass expanding the external mandibular cortex from the 2 deciduous molar to the 1 permanent molar (arrow).

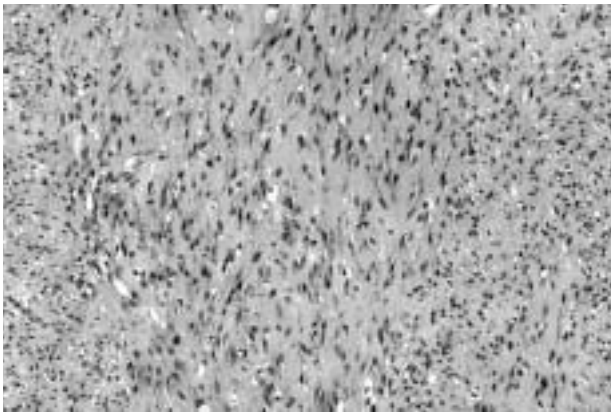

Figure 2. Appearance of the neoplasm showing interlacing fascicles of spindle-shaped tumor cells. Bar is 50 microns, H&E (arrow).

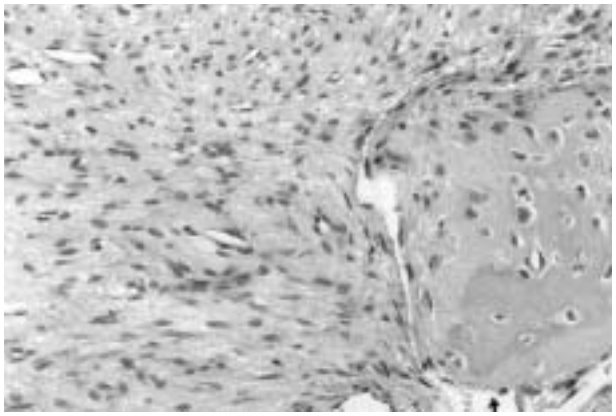

Figure 3. The tumor consisted of interlacing bundles of spindles cells with uniform cigar-shaped nuclei, lacking significant mitotic activity. A bone spicule, on the right, is surrounded by neoplastic cells. Bar is 25 microns, H&E (arrow).

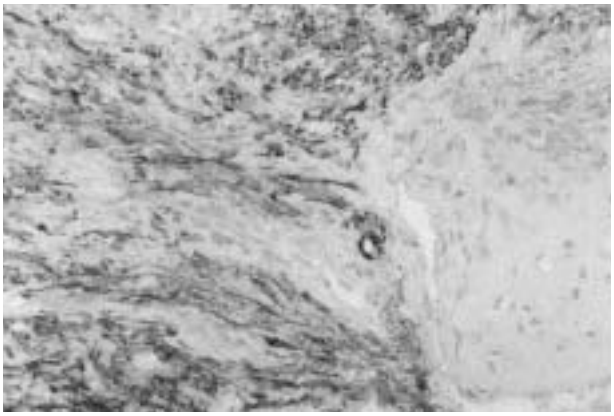

Figure 4. The same microscopic field of Figure 3 shows a strong immuno-histochemical positivity for smooth muscle actin. Bar is 25 microns.

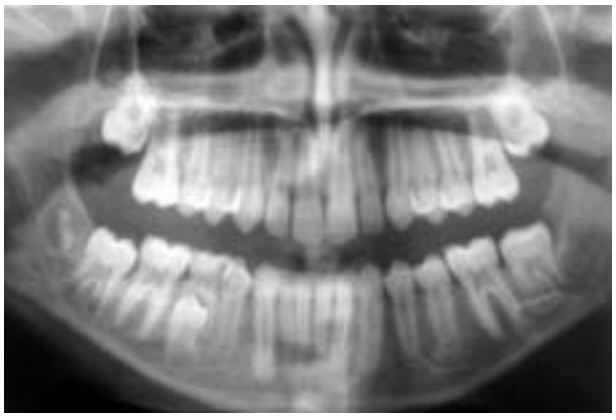

Figure 5. Panoramic radiograph 40 months after surgery. There are no signs of clinical and radiographic recurrence. The permanent germs on the left side of the mandible were preserved and are erupted. Only the third molar germ was excised.

The histological examination showed an expansile neoplasm with bone at the periphery. The tumor consisted of interlacing bundles of spindle cells with uniform cigar-shaped nuclei, lacking significant mitotic activity (Figure 3). Immunochemistry demonstrated that the spindle-shaped cells were mainly smooth muscle cells, with occasional collagen fibers interspersed, staining strongly for smooth muscles markers (Figure 4). A diagnosis of benign leiomyoma was made.

Postoperatively, progress was unremarkable and after three years there is no evidence of recurrence. The panoramic radiograph at the last follow-up control (November 2002) showed normal permanent tooth eruption (Figure 5).

DISCUSSION

Mandibular leiomyoma is a rare benign tumor, accounting for less than 0.1% of all cases occurring in the oral cavity.[3] The rarity of the lesion could be explained by the small amount of smooth muscle in the

mandible. The possible origin has been discussed by many authors.[4-6] The vascular walls may be the only source of smooth muscle, but heterotopic embryonal tissue also has been suggested.[3]

The lesion is described as slow-growing, with no sex predilection, and an average-age of presentation of 42.7 years.[5] In half of the previously reported cases of leiomyoma in the mandible, the tendency for rapid growth is recognized.[7] The leiomyoma in this patient also showed a quite rapid increase in size, even correlated with the mandibular and general growth of the child. Normally, the differential diagnosis of vascular leiomyoma in the body of the mandible includes vascular lesions such as central giant cell granulomas, hemangiopericytomas, traumatic bone cysts, arteriovenus malformation, and odontogenic cyst. In case of intraosseuos leiomyoma such as the one described, the panoramic radiograph could not reveal any radiolucency, or only a unilocular image, even if some cases described of intrabony tumors presented as multilocular radiolucencies.[8,9]

Sometimes, only the CT scan examination shows the lesion, which generally appears subcortical in origin. This particular radiographical appearance suggests a differential diagnosis with connective tissue origin.

Three types of benign smooth muscle tumors are described by Enzinger *et al.* in 2001: leiomyoma, angiomyoma (vascular leiomyoma), and epitheliod leiomyoma (bizarre leiomyoma).[10] The case presented here is an example of non-vascular leiomyoma, similar to that presented by McLeod *et al*.[8] and Raffaini *et al*.[1]

The treatment of choice is surgery. In the case reported, the local excision via intraoral approach was justifiable in view of the benign nature of the tumors, avoiding visible scar on the face of the young patient.

Recurrence after adequate surgical treatment is unusual, although it has been reported in the vascular type.[11,12]

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