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CASE REPORT

Intraosseous leiomyoma of the mandible

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Summary The case of an intraosseous leiomyoma in a 57-year-old man is presented. The tumor was incidentally discovered during routine dental X-ray examination as a round, unilocular, radiolucency, measuring approximately 2×1.5 cm. Clinical examination revealed a slight swelling of the buccal cortical plate in the edentulous area of the posterior right mandible, and intra-operatively, perforation of the buccal cortical plate was seen. Histological and immunohistochemical examination was diagnostic of solid leiomyoma. One year after surgery the patient is free of residual or recurrent disease. This is the fifteen documented cases of intraosseous leiomyoma reported in the English literature.

Introduction

The leiomyoma is a benign smooth-muscle neoplasm commonly found in the female genital tract, gastrointestinal tract, or skin. Leiomyomas of the oral cavity are unusual, with less than 125 documented cases reported up to 1998.[1] The lips, palate, tongue, and gingiva are the intraoral sites where leiomyomas more commonly occur.[2] Intraosseous leiomyomas of the jaws, thought to originate from vascular smooth muscles or pericytes,[3] are extremely rare. Since the recent review of 13 cases by Liang et al.,[4] only one more case has been reported.[5]

We present the case of a solid intraosseous leiomyoma in the posterior mandible of a 57-year-old man.

Case report

A 57-year-old Caucasian man was referred for a well-defined mandibular radiolucency, incidentally discovered by his general dental practitioner during routine dental X-ray examination. The patient did not report pain or sensory alteration in the area. His medical history was non-contributory.

Clinical examination revealed a slightly compressible swelling of the buccal cortical plate in the edentulous area of the posterior right mandible, covered by intact mucosa.

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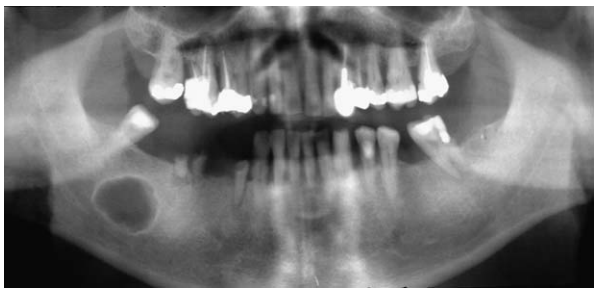

Figure 1 The panoramic radiograph shows a round, unilocular, radiolucency with sclerotic margins, in the right side of the mandible, in contact with the inferior alveolar nerve canal.

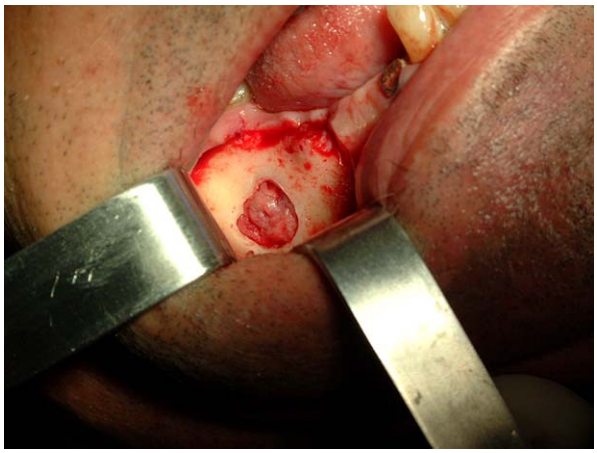

Figure 2 The tumor appears as a solid mass through the perforation of the buccal cortical plate.

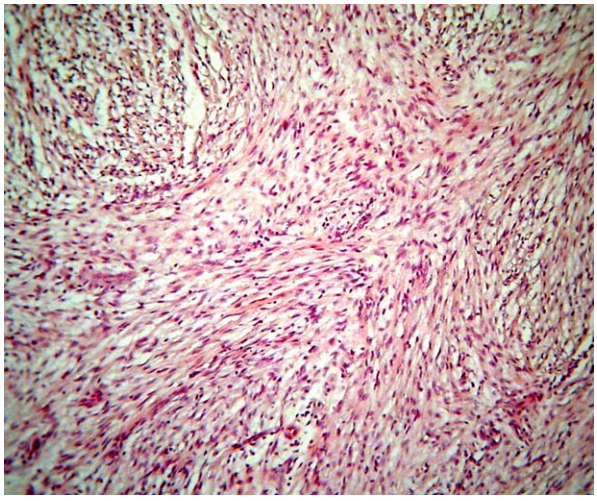

Figure 3 Interlacing fascicles of spindle shaped cells with a single, pale staining, tapered nuclei, and eosinophilic cytoplasm (hematoxylin and eosin stain, original magnification ×40).

It was asymptomatic on palpation and the adjacent teeth did not show pathologic mobility. Cervical examination was within normal limits.

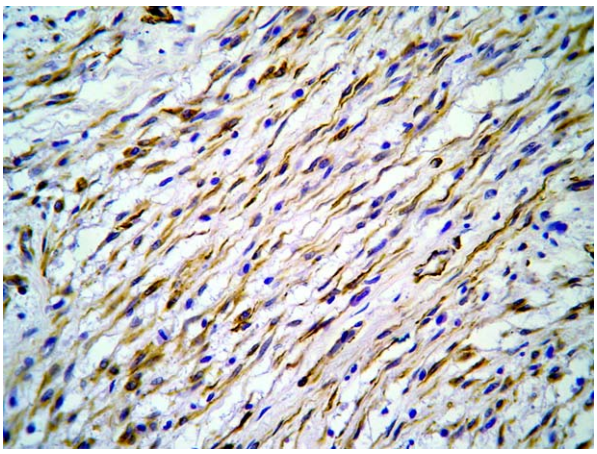

Figure 4 Intense α-smooth-muscle actin positivity of the tumor cells (α-smooth-muscle actin, original magnification ×100).

The panoramic radiograph showed a round, unilocular, radiolucency with sclerotic margins, measuring approximately 2 × 1.5 cm (Fig. 1). It was in contact with the inferior alveolar nerve canal, and was not associated with the roots of the adjacent teeth.

With the presumptive diagnosis of an odontogenic cyst or tumor, total excision of the lesion through an intraoral approach was planned. Upon reflection of the flap, a perforation of the buccal cortical plate was revealed (Fig. 2). The lesion appeared as a solid mass that was easily enucleated through the widened cortical perforation.

Five micron thick formalin-fixed and paraffin-embedded tissue sections showed a well-circumscribed, fascicular proliferation of spindle shaped cells with a single, pale staining, tapered nucleus and eosinophilic cytoplasm (Fig. 3). No nuclear or cellular atypia were evident and mitotic figures were scant. Immunohistochemically, the tumor cells were positive for vimentin and α-smooth-muscle actin (Fig. 4), focally positive for desmin, and negative for S-100 protein. The final diagnosis was solid leiomyoma.

One year after surgery, the patient is free of residual or recurrent disease and a radiographic examination showed normal bone regeneration.

Discussion

Fifteen cases of intraosseous leiomyoma of the jaws, including the present one, have been reported in the literature.[4,5] Thirteen cases occurred in the mandible, mostly the posterior region. The age of the patients ranged from 8 months to 71 years, and the average age at onset of the 15 cases was 35.4 years. Eleven of 15 patients were Caucasians, with a 9:5 male predominance. Duration of the lesions varied from 3 weeks to 2 years. Most lesions presented as swellings, but in three patients, including the present one, the tumor was an incidental radiographic finding. The case of Brooks et al.[2] is unusual in that it manifested as an endodontic/periodontic lesion associated with a vital mandibular lateral incisor.

Most lesions were described as unilocular or multilocular radiolucencies measuring more than 2 cm, with well- or ill-defined border. Root resorption and erosion of the alveolar ridge or the cortical plates were common findings. It should

be mentioned, however, that large dimensions, rapid enlargement, and loss of the cortical bone may be suggestive of malignancy.[1] Due to the lack of diagnostic features, the differential diagnosis includes more common intraosseous lesions, such as central giant cell lesion, ameloblastoma, myxoma, traumatic bone cyst, hemangioma, and neurofibroma.[6]

Microscopically, eight cases were solid leiomyomas and six vascular leiomyomas or angiomyomas. In one case[5] the subtype was not stated, but the description and the figures are consistent with a solid leiomyoma. Immunohistochemistry is necessary for arriving at the final diagnosis, as the more common solitary myofibroma of the jaws may have a similar microscopic appearance and is actin-positive, but is also desmin-negative.[7] In addition, the solitary myofibroma occurs almost exclusively in the mandible of patients under 18 years of age.[7] Differentiation of leiomyoma from leiomyosarcoma is not always easy because there are no clear-cut diagnostic criteria for malignancy in a leiomyoma.[1]

Intraosseous leiomyoma is a benign tumor treated by surgical excision. Complete conservative removal is curative, and no recurrence was reported in 13 patients with follow-up, 9 months to 11 years post-operatively.

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