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CASE REPORT

A very rare form of leiomyoma: Mandibular angioleiomyoma

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Summary Mandibular angioleiomyomas are extremely exceptional, and they may stem from the smooth muscle of vessel walls, aberrant adnexial smooth muscle, arteriovenous anastomoses, ectopic thyroglossal ducts and hamartomas. In this report, a case of angioleiomyoma in the posterior mandibular area was presented, and aetiological, clinical, radiographical, histological characteristics and treatment modalities of the lesion were discussed.

1. Introduction

Leiomyomas are benign, smooth muscle neoplasms that may emerge from aberrant adnexial smooth muscle cells [1], or their precursors in the media of blood vessels, in the muscularis layer of the gut, and in the body of the uterus [1—5]. According to World Health Organization, leiomyomas are classified in three histological groups [6—9]: (a) vascular [angioleiomyomas], 74% of the cases; (b) solid, 25% of the cases; (c) epitheloid [leiomyoblastomas], less than 1% of the cases [8].

Leiomyomas of the oral cavity are rare due to the lack of smooth muscles within the oral cavity [4,7,10], and those of the mandible are extremely exceptional [1]. Among the origins of oral leiomyomas smooth muscle of vessel walls, the circumvallate papilla, and atypical arrectores pilorum muscles in the cheeks are cited [6].

The number of oral leiomyoma cases vary in the literature between 4 and 125 [1,2,4,6,10,11]. Clinically, they are slow-growing, generally asymptomatic submucosal masses [4,9,10,12—14], which can be seen at any age ranging from infancy to 76 years [9], but mostly in the 40—59 year group [4,11], and are usually discovered when they are 1—2 cm in diameter [5]. Although rarely, difficulty in chewing, swallowing and opening the mouth, a change of voice [2], and occasional periods of numbness [13] are reported. The predilection sites are lip (27.46%), tongue (18.30%), palate, and buccal mucosa (15.49%) [4,11,12,16,15]; however, every now and then, the first two sites are swapped in some reports

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[5,7,9]. The tumor has also been detected in gingiva [4,11,12,15,16], mandible [15,16], floor of the mouth [4,11], salivary glands [4,17], uvula [4], zygoma [4], palate [6,14], nasal cavity [3,14], and maxillary tooth socket [4].

Clinically, 55.9% of the angioleiomyomas appear as red lesions; the rest are mostly gray, white or the color of normal mucosa [11]. Radiographically, leiomyoma may cause advanced bone loss [6,12], consequently leading to tooth mobility [6].

2. Case report

Twelve-year-old Caucasian male was admitted to the Department of Oral Diagnosis and Radiology, School of Dentistry, Ege University with the complaint of swelling in the left retromolar region. His medical history was non-contributory, and his family had no significant medical problems. Clinical examination revealed a rubbery mass of 2 cm in diameter in left mandibular third molar area on the alveolar crest, with a small ulcer on its surface (Fig. 1). The second molar was located in a vestibular position. The regional lymph nodes were normal.

Radiographic examination on the orthopantomographic film disclosed a radiolucent mass in the left retromolar area, affecting the lingual compact bone. Additionally, mandibular third molar's angulation and localization were changed due to the pressure of the mass (Fig. 2). In three dimensional computer tomographic images [3D-CT] and conventional CT, a mass extending down lingually to the mylohyoid line, compressing the lingual plate of the mandibular compact bone was observed (Fig. 3).

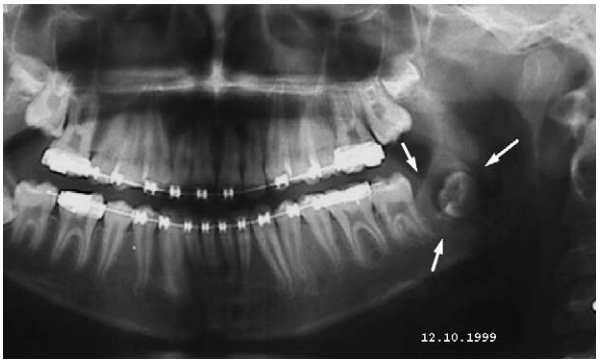

Fig. 2 Orthopantomograph of the patient prior to the operation.

On 26 October 1999, an incisional biopsy was performed under local anesthesia, and a biopsy specimen that was a vascular appearing soft tissue, approximately 0.5 mm in diameter was retrieved. The biopsy report affirmed that the lesion was an angioleiomyoma, and total excision of the lesion was planned. Meanwhile, after taking the biopsy specimen, the lesion had expanded laterally and lingually on the alveolar crest (Fig. 4).

The surgical intervention was performed under local anesthesia on 9 November 1999. After administration of 3% citanest-octapressin, an incision with approximately 3 mm security demarcation lines was made on the vestibule of the retromolar area passing through the normal gingiva. An additional incision distal to the first molar was created, and a mucoperiosteal flap was elevated. A silk suture was used to hold and raise the mass from the alveolar crest, and its association with mandible was eradicated by curetting the mass with a raspatorium.

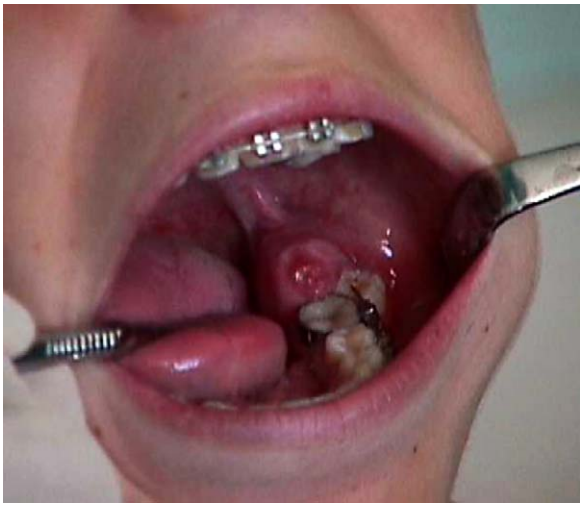

Fig. 1 Intraoral view of the patient prior to the biopsy, revealing a small ulcer on the mass and displacement of the second molar.

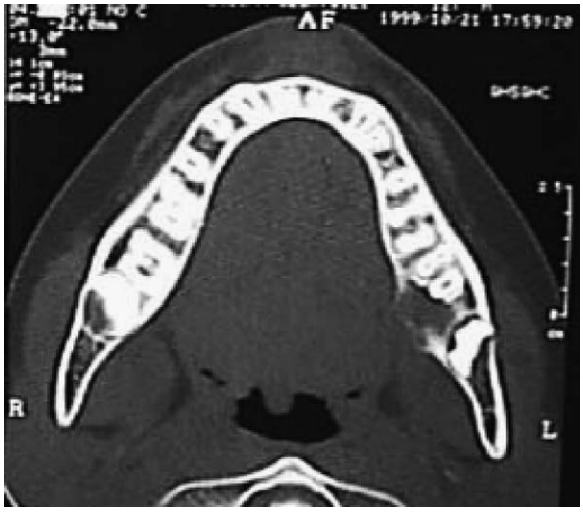

Fig. 3 Conventional CT image of the patient clearly demonstrating the lesion, compressing the lingual plate of the mandibular compact bone.