Oral leiomyoma in retromolar trigone. A case report

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ABSTRACT
Leiomyomas are benign tumors originated on smooth muscle. The most frequent site of appearance are uterine myometrium, gastrointestinal tract and skin. The highest incidence occurs between 40 and 49 years of age. Its presentation is unusual in children or in older patients. Leiomyomas are infrequent in the oral cavity, but in this location are usually localized on tongue, lips and palate.
Leiomyomas use to appear as well-defined masses, with slow growth and totally asymptomatic. Pain is present just in rare cases. The treatment is surgical excision. Recurrences are extremely infrequent.
The diagnosis is mainly determined by histological studies due to its unspecific clinical appearance. Histopathologically proliferation of smooth muscle cells is observed without necrotic areas. A low number of mitotic figures can be seen.
We present the case report of a 25-year old male patient, with a leiomyoma on his right retromolar trigone. The low incidence of this pathology, the age of the patient and the unusual location, make the report of the case worthy.

Key words: Leiomyoma, oral cavity, oral tumour.

RESUMEN
Los leiomiomas son tumores benignos originados en el músculo liso. Su localización más frecuente es el útero, el tracto gastrointestinal y la piel. La incidencia más alta ocurre entre los 40 y 49 años de edad, siendo muy rara su aparición en la infancia y en la senectud. Son muy infrecuentes a nivel de la cavidad oral, pero cuando se dan en esa localización, asientan principalmente en la lengua, los labios o el paladar.
Inicialmente suelen presentarse como una masa muy bien definida, de lento crecimiento y totalmente asintomática. En raras ocasiones producen dolor. Su tratamiento es casi siempre quirúrgico, siendo las recurrencias excepcionales.
Dada su clínica inespecífica, su diagnóstico es principalmente histológico, observándose en las muestras una proliferación de células musculares lisas, sin focos de necrosis y con escasas mitosis.
Presentamos el caso clínico de un paciente de 25 años de edad con un leiomioma en trigono retromolar derecho. Dada la escasa incidencia de este tipo de patología, la edad del paciente y su inusual ubicación, se justifica la presentación de este caso.

Palabras clave: Leiomioma, cavidad bucal, tumor oral.
INTRODUCTION
Oral leiomyoma has a low incidence. When it presents on this location, it appears as a slow-growth mass, symptomatic just in exceptional cases (1). Main symptoms are pain, teeth mobility, or even difficulty in chewing, usually when the tumour is located in tongue, lips or palate (2).
Due to its unspecific clinical presentation, diagnosis is made after histological study (3), where the typical small smooth muscle cells are seen, with uniform-size and with no malignity criteria. In order to achieve more specific analysis, and more precise differential diagnosis, it’s possible to make immunohistochemical studies.
Surgery is the only successful treatment at this moment. It’s important to obtain a complete resection in order to avoid recurrences (2) (usually easily due to its characteristics as a well-circumscribed tumour).

CASE REPORT
A 25-year-old male patient, without remarkable medical history or toxic habits, came to our clinic with a 1 x 1.5 cm tumor placed on right retromolar trigone. The mass had appeared three weeks before. It was painless, no adhered and well-defined with normal mucosa overlying (Figure 1). Fine Needle Aspiration was negative for malignant cells. The result was unspecific with a possible glandular origin. Semi-inclusion of the teeth 4.8 was observed, with no apparent relation with the lesion. No palpable cervical nodes were found. CT scan of the neck showed bilateral 1 cm. lymph nodes on level II, and subcentimetric ones on right level III and on submaxilar area. No masses or pathological contrast captation were observed.
The mass was excised under general anaesthesia with a 1cm free-margin. The diagnosis suspected was a lesion originated on minor salivary glands of retromolar trigone.
The patient had a correct evolution without any post-operative incident.
Macroscopically the tumor had greyish colour, elastic consistence, rough appearance and it was encapsulated (Figure 2)
The histological report informed that the tumour was constituted of small spindle cells, with eosinophilic cytoplasm and central and basophilic nucleus. The cells appeared in a typical whirling pattern. The lesion showed prominent vascular spaces between the bands of smooth muscle cells. No mitotic figure, nuclear atypia or necrotic areas were identified. Histological diagnosis was leiomyoma, with a clear origin on the smooth muscle placed under oral mucous (Figure 3).
The patient is disease free one year after surgical excision.

DISCUSSION
Leiomyoma is a benign tumour of smooth muscle. It can appear in any location where this kind of tissue is present. This lesion occurs most often in female tract. This represents approximately 95% of the total (4). The remaining 5% use to be placed in gastro-intestinal tract and skin. Oral leiomyomas are considered uncommon neoplasms, around 0.065 %.
The origin of leiomyomas of oral cavity is restricted to three areas with smooth muscle in their histological analysis: tunica media of the blood vessels (5), ductus lingualis and circumvallate papilla (6).

Since 1884(7), when oral leiomyoma was first reported, less than 150 cases have been published (8).

The most common sites of occurrence of the oral leiomyoma are tongue, lips, palate and cheek (2). Other less frequent locations are floor of mouth and gingiva. Retromolar trigone is an unusual place, according to the reviewed literature.

Clinically, the leiomyomas use to be unspecific masses, with several aspects from normal to more congestive mucosa, according to their vascularity (9). These use to be well-defined, slow growing, asymptomatic masses. Some symptoms are induced (1) due to local growth. Deglutition difficulty, toothache, loose teeth, or pain refered in TMJ (10) have been reported. Shortness of breath can be produced by huge tumours (11). Pain is suspected to be prompted by local ischemia due to tumoral vessel contraction (12) and to neural irritation near the tumour (13).

Oral leiomyoma is more frequent in men than in women, with a 1.43:1 ratio (9). Peak age incidence is 40-49 years old (2).

There have not been risk factors related with this kind of tumour. In our case, there is not suspected relation between semi-erupted teeth 4.8 and local tissue damage and the leiomyoma.

Several oral tumours should be included in the diferencial diagnosis, benign lesions such as fibroma, neurofibroma, lipoma or mucocele or malignant ones such as leiomyosarcoma. Histological studies are necessary to achieve diferencial diagnosis.

The definitive diagnosis in leiomyoma is the histological one. Typical smooth muscle cell proliferation (small and spindle) is observed without necrosis areas or mitotic figures. The malignant counterpart: the leiomyosarcoma should be taken into account when the number of mitotic figures per field is over ten (14). Difference between leiomyoma and low grade leiomyosarcoma is not always easy (15).

Histologically three types of leiomyomas can be differentiated (3): solid leiomyoma (25 %), vascular leiomyoma or angiomyma (74 %) and leioblastoma or ephiteoid leiomyoma (1 %).

Hematoxylin and eosin stains can be used in this kind of tumour. Special stains such as Masson’s trichrome, Van Gieson’s stain, or Mallory’s phosphotungstic acid (PTAH) are specific for muscle cells and collagen fibres. Immuno-histochemical detection of actine (smooth muscle marker) can also be useful.

Wide surgical resection is the only reported treatment in reviewed literature with successful results. Recurrence rate is very low if complete resection is achieved (2). It has been reported a recurrence case two weeks after surgery (16), probably due to incomplete resection.

REFERENCES