Unusual oral leiomyoma in a child: case report

**ABSTRACT**

**Aim** Very few oral leiomyomas have been reported in the literature, especially in children. The aim of this article was to present a very rare new case of leiomyoma in the chin, in a seven-year-old black male, and to explain its clinical and histological features.

**Case report** A seven-year-old black male patient presented with a swelling mass of diameter 2.5 x 3.0 cm in the lower anterior area of the chin. The tumor was firm, painless, non-fixed and well-defined, with normal mucosa overlying it. Fine-needle aspiration prior to surgery was negative. The surgical access was intraoral and the mass was located under the skin and below the periosteal bone, within the musculature. The histological and immunohistochemical studies diagnosed an oral leiomyoma. The clinical appearance of leiomyomas is very difficult to differentiate from other mesenchymal tumors. Histological and immunohistochemical studies were crucial for achieving a definitive diagnosis. Eighteen months after the excision of the lesion, no signs of recurrence were observed. The successful treatment had several objectives, including removing the lesion with success and providing the patient with an aesthetic appearance.

**Keywords**: Leiomyoma; Oral; Child.

Introduction

Leiomyoma is a benign smooth-muscle tumor that can appear in any location. The most frequent sites are the female genital tract, gastrointestinal tract and skin. However, it is very rare in the oral cavity [Baden et al., 1994]. There are three types of leiomyoma according to the World Health Organization classification: solid leiomyomas, vascular leiomyomas (angiomyoma) and epitheloid leiomyomas (leiomyoblastoma). The solid and vascular types are the most frequent variants [Baden et al., 1994].

According to Farman [1975], the prevalence of smooth-muscle tumors at intraoral locations is only 0.065%. This is because of the lack of smooth muscle in this part of the body. A review of the literature found only five cases of oral leiomyoma in a series of 7,748 leiomyomas from all sites [Baden et al., 1994].

Oral leiomyoma can appear at any age. However, the greatest prevalence is in the age group between 40 and 49 years old, and it is very rare in children [Epivatianos et al., 1985; Wertheimer-Hatch et al., 2000].

The most frequent locations for these rare oral leiomyomas are the lips, tongue, hard and soft palate and, much less frequently, the cheeks [Epivatianos et al., 1985; Wertheimer-Hatch et al., 2000]. Few authors have reported leiomyomas located in unusual oral sites like the mandible [Kabou et al., 1997; Rey et al., 2007] and zygomatic bone [Robiony et al., 1996].

Clinically, when the tumor is present, it generally occurs as a slow-growing mass, presenting as a firm mucosal nodule. Most lesions are asymptomatic, although occasionally they may be painful [Cherrick et al., 1973; Orsini et al., 2001].

Surgical excision is the recommended treatment for leiomyoma, with a high success rate. The risk of recurrence is low, if complete resection is achieved [Orsini et al., 2001].

The diagnosis of leiomyoma in the oral cavity is mainly determined by histopathological studies to confirm its smooth-muscle origin. Immunohistochemical studies may be helpful to differentiate leiomyoma from other tumors. Proliferation of smooth-muscle cells presenting uniform size without necrotic areas or any criteria for malignity is observed. A low number of mitotic figures may be seen [Rey et al., 2007; Ezinger et al., 1969].

Very few oral leiomyomas have been reported in the literature, especially in children. The purpose of this article was to present a very rare new case of leiomyoma in the soft chin, in a seven-year-old black male, and to explain its clinical and histological features.

**Case report**

A seven-year-old black male patient without any remarkable medical history came to the Oral and Maxillofacial Department of São Leopoldo Mandic Dental Research Institute with a history of swelling in the soft chin region. His mother reported that the mass had appeared about three months earlier and that, since then, it had been growing slowly and continuously (Fig. 1). The patient was not taking any medications.

Clinically, a swelling mass of diameter 2.5 x 3.0 cm was found in the lower anterior area of the chin. The tumor was solid to touch, painless, non-adherent and well-defined, with normal mucosa overlying it. There were no signs of infection or fistula and the local temperature was normal. Intraoral examination did not show any abnormality in the overlying mucosa. No palpable cervical nodes were found. Orthopantomography and periapical X-ray and occlusal imaging were requested. However, there were no pathological findings from these.

The lesion was excised under local anaesthesia and nitrous oxide sedation. Fine needle aspiration prior to surgery was negative. The surgical access was intraoral. The well-defined mass, located between the periosteum and the...
skin, was removed and the incision site was sutured (Fig. 2).
The patient had good and correct evolution without any postoperative incident.
The histological report stated that the tumor was a well-delimited solid cellular mass consisting of small fusiform cells with uniform monochromatic nuclei and blunt-ended spindles. The cells were arranged in intersecting fascicles and no pleomorphism, mitotic activity, nuclear atypia or necrotic areas were found (Fig. 3A). The immunohistochemical study revealed the expression of alpha-smooth muscle actin (Alfa-SMA), that is commonly used as a marker of myofibroblast formation (Fig. 3B). Based on all these findings, a diagnosis of oral leiomyoma was made.

Thirty-one months after the excision of the solid leiomyoma, no signs of recurrence were observed. The soft tissues appeared healthy. The successful treatment had several objectives, including providing the patient with an aesthetic appearance and removing the lesion with success.

Discussion

Leiomyoma is a relatively uncommon lesion on the face and in the oral cavity because of the scarcity of smooth muscle. The possible origins of the leiomyoma in these sites are restricted to three areas that have smooth muscle: the tunica media of the blood vessels [Stout, 1937], the ductus lingualis and the circumvallate papillae [Glas, 1905].

In 1884, Blanc reported the first case of leiomyoma in the oral cavity. Since then, very few cases of oral leiomyoma have been described in the literature [Orsini et al., 2001; Benet et al., 2003]. From reviewing the literature, it can be seen that the soft chin region is definitely an unusual place to find a leiomyoma, especially in a seven-year-old child, since the age group with the highest incidence of leiomyomas is between 40 and 59 years [Epivatianos et al., 1985; Wertheimer-Hatch et al., 2000].

Out of all of the cases of oral leiomyoma described in the literature, the youngest patient was a six-day-old infant who had a leiomyoma at the base of the tongue and required intubation at birth for airway obstruction [Kotler et al., 1994]. This present report adds a new rare case to the literature on leiomyomas in children.

The majority of the reported cases have indicated that leiomyomas are asymptomatic, slow-growing masses [Cherrick et al., 1973; Benet et al., 2003]. The case described here is in agreement with the recent literature.

Our seven-year-old patient did not report any discomfort during the period when the tumor was growing. Even though the excised tumor could have caused pain, tooth mobility or even difficulty in chewing [Wertheimer-Hatch et al., 2000; Orsini et al., 2001], no such symptoms were reported. This may have been because such symptoms are more often related to tumors growing in the lips, tongue or palate.
When pain occurs, it is suspected that it is prompted by local ischaemia due to tumor vessel contraction [Duhing and Ayer, 1959] and neural irritation near to the tumor [Toida, 2000]. These are not very big tumors, with an average size of 1-2 cm and with a history of less than one year of evolution [Epivatianos et al., 1985]. This leiomyoma in the chin was slightly bigger than the average (2.5 x 3.0 cm) and had a history of only three months, which means that the excised leiomyoma was a large tumor that had grown over a short period of time.

The clinical appearance of leiomyomas is very difficult to differentiate from other mesenchymal tumors. The differential diagnosis must include several oral tumors: benign lesions such as fibroma, neurofibroma, lipoma, mucocele, focal epithelial hyperplasia, rhabdomyoma, schwannoma or pyogenic granuloma; and malignant ones such as leiomyosarcoma, fibrosarcoma or rhabdomyosarcoma [Rey et al., 2007; Benet et al., 2003].

The histological analysis is crucial for achieving a definitive diagnosis. A diagnosis of a typical leiomyoma can normally be made from the macroscopic findings and observation using hematoxylin and eosin staining. Special staining, such as Masson’s trichrome stain can be useful for demonstrating collagen fibers between smooth-muscle cells and myofilaments within them [Grippaudo and Becelli, 1996]. In our case, only hematoxylin and eosin staining was performed.

We undertook a complementary histological study using the immunohistochemical technique, and this was positive for the smooth-muscle markers MSA and SMA. For differential diagnosis, S-100 protein and CD34 were used, but both of these were negative. These results corroborate the literature [Rey et al., 2007; Ezinger et al., 1969]. In order to differentiate the leiomyoma from low-grade leiomyosarcoma, we investigated the number of mitotic figures per field. Fewer than two mitotic figures per 10 high-power fields generally indicates a good prognosis, and no mitotic figures were found in our case [Gorlin and Goldman, 1970].

In order to avoid recurrences, it is important to achieve complete resection during the operation, as was done in our case when the tumor in the chin was removed. This procedure is usually easy to perform because of the well-delimited nature of this type of tumor [Wertheimer-Hatch et al., 2000].

Conclusion

The clinical appearance of leiomyomas is very difficult to differentiate from other mesenchymal tumors. Histological and immunohistochemical studies were crucial for achieving a definitive diagnosis. The successful treatment had several objectives, including removing the lesion and providing the patient with an aesthetic appearance.

References


