

# LEIOMYOMA OF THE MANDIBLE IN A 9YEAR OLD CHILD

## - A CASE REPORT

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## **Abstract**

Leiomyomas are the benign tumours of the smooth muscle that usually arise in the gastrointestinal system and in the uterus. Oral Leiomyomas are uncommon due to the paucity of the smooth muscles in the mouth (**except in blood vessels**) and those of the mandible are extremely rare. Leiomyomas have been classified as solid, angiomyoma (vascular leiomyoma), and epithelioid variants. Here, we report a rare case of leiomyoma of the mandible in a 9year old child together with conventional histopathologic and immunohistochemical findings.

Keywords: leiomyoma, smooth muscle tumor, immunohistochemistry

## **Introduction**

Leiomyoma was first reported by Blanc in 1884<sup>1</sup>. Leiomyoma is a benign tumour of smooth muscle origin which is usually diagnosed in the gastrointestinal tract, uterus and skin.<sup>1, 2, 3, 4</sup> It most commonly arises in the retroperitoneum, mesentery, omentum or subcutaneous and deep tissues of the limbs<sup>5</sup>. It is found uncommonly in the oral cavity because of the paucity of the smooth muscles in this region (except in the blood vessel walls)<sup>1</sup>. If at all it occurs, it shows its predilection in the posterior portion of the tongue, lips, palate, cheeks, gingiva and salivary glands<sup>6</sup>. Intraosseous oral leiomyomas are rare and those of mandible are very rare<sup>3</sup>. The origin of oral leiomyomas is smooth muscle of vessel walls, the circumvallate papilla and atypical arrectores pilorum muscles in the cheek<sup>7</sup>. Clinically, they can be seen at any age ranging from infancy to 76 yrs, but mostly seen in middle aged group<sup>6, 7</sup> and exhibits male predilection<sup>8</sup>. Radiographically, angioleiomyomas manifest as unilocular or multilocular radiolucent lesions with either an ill-defined or a well-defined sclerotic border. Cortical expansion of the alveolar plates may be evident. Root resorption has also been shown<sup>4</sup>. Histopathologically, it is composed of spindle cells arranged in whorled and interlaced fascicular pattern. The cells show elongated nuclei with fusiform or blunt ends<sup>2</sup>. The line of treatment is conservative surgical excision<sup>1, 2</sup>.

## **Case Report**

A 9year old male child reported to the Department of Oral & Maxillofacial Pathology, SIBAR Institute of Dental sciences with an asymptomatic extra oral swelling on right body of the mandible that had been evident for 2 months [Fig 1]. According to the patient, the swelling was initially small and gradually progressed to the present size and was asymptomatic. His past medical history and family history were non contributory. On clinical

examination, right facial asymmetry was observed. A solitary, extra oral swelling was seen on the right side of the body of the mandible which was oval in shape and was 4x3 ½ cm in size extending anteriorly 2cm away from the corner of the mouth and posteriorly 1cm in front of the angle of the mandible. Superiorly, extending up to the line joining the corner of the mouth and the tragus of the ear and inferiorly to the lower border of the mandible. **It was hard and non tender on palpation** [fig 1]. Intraorally, the overlying mucosa was intact and smooth and there was no tooth mobility. The regional lymph nodes were not enlarged. **The clinical differential diagnosis includes central ossifying fibroma, central giant cell granuloma, neurofibroma, neurilemmoma, myofibroma of the mandible and osteoma were considered.**

On radiographic investigations, the panoramic radiograph of the right mandibular premolar –molar region showed faint periosteal reaction in the vicinity of permanent first molar. The developing tooth buds of right canine and premolars appear to be uninfluenced by the pathosis in the region as compared to the left side. The periosteal bone reaction is noted (fig-2). As the panoramic radiograph revealed no significant findings an axial CT was advised. The axial CT view in bone window at the level of inferior border of the mandible showed a massive periosteal reaction on the buccal surface demonstrating the sclerotic borders of the lesion. The lesion appears to have soft tissue density within the central zone as seen from this window suggesting that the lesion is of soft tissue region. (Fig-3). A radiographic differential diagnosis of neurofibroma, neurilemmoma, myofibroma, desmoplastic fibroma of the mandible, adenomatoid odontogenic tumor, periosteal osteoblastoma and periosteal osteosarcoma were suggested. Clinico-radiographically, a provisional diagnosis of myofibroma of the mandible was given.

The lesion was removed through the use of extra oral surgical approach with the patient under general anaesthesia. A well circumscribed lesion was

observed and the lesion was easily removed from the surrounding tissues. Macroscopically, the lesion was roughly a spherical mass, brownish white in color measuring 1x1x1 cm and was firm in consistency. Microscopically, the tissue section exhibited cellular areas intermingled with collagenous and hyalinized areas. The cells were predominantly spindle shaped and showed elongated nuclei with fusiform or blunt ends [Fig -4]. The tissue also exhibited numerous vascular areas. In some areas the cells along with the collagen fibers were arranged in fascicles. The periphery of the lesion exhibited spicules of vital reactive bone. Most of the trabeculae were perpendicularly arranged with respect to the periphery of the lesion. There was a minimal diffuse chronic inflammatory cell infiltrate. **Histopathologically, a diagnosis of spindle cell neoplasm was given.**

**All the spindle cell neoplasms show similar histopathological features, so Immunohistochemistry was suggested.** IHC was performed against vimentin, smooth muscle actin and S-100 protein. The tumour was positive for  $\alpha$ -smooth muscle actin [Fig-5] and vimentin [Fig -6] and negative for S-100 protein and desmin . **Based on the IHC report a confirmatory diagnosis of leiomyoma was given.** No complications were observed during the consecutive post operative sessions in past three years and a perfect bone healing occurred without any recurring pathoses.

## **Discussion**

The occurrence of leiomyoma is rare in the oral cavity. It accounts for 0.42% of all soft tissue lesions in the oral cavity<sup>9</sup>. The intraosseous leiomyomas are very rare<sup>1, 6, 9, 10</sup>. A review of the English language literature showed that 10 cases have been described<sup>1</sup>.

Huseyin Koca et al reported that they can be seen at any age ranging from infancy to 75 years and are most commonly seen in the middle aged group<sup>6, 7</sup>. It

exhibits slight predilection for males<sup>8</sup>. The most common sites of oral leiomyoma are posterior portion of the tongue, lips, palate, cheeks, gingiva and salivary glands<sup>6</sup>. Intraosseous lesions are rare and if at all they occur show predilection for mandibular posterior region with cortical involvement<sup>11</sup>. The rarity in the mandible could be explained by the small amount of smooth muscle in the mandible. The vascular walls may be the only source of smooth muscle in the mandible, but heterotopic embryonal tissue also has been suggested<sup>1, 3</sup>. In our case the age, gender and site predilection were consistent with the earlier reports.

Radiographically, angioleiomyomas manifest as unilocular or multilocular radiolucent lesions with either an ill-defined or a well-defined sclerotic border. Cortical expansion of the alveolar plates may be evident. Root resorption has also been shown<sup>4</sup>. In our case the panoramic radiograph revealed no characteristic findings and hence an axial CT was advised.

According to World Health Organization, leiomyomas are classified into three histological groups, (a) Vascular [angioleiomyoma], accounts for 74% of the cases; (b) solid, accounts for 25% of the cases; (c) epithelioid [leiomyoblastomas], accounts for less than 1% of the cases<sup>5,10</sup>. Histologically, the tumour is composed of spindle cells arranged in a whorled and interlaced fascicular pattern. The cells showed elongated nuclei with fusiform or blunt ends, and perinuclear vacuolization can sometimes be noted. Mature smooth muscle cells have the distinctive characteristics of small and uniform nucleus and broad eosinophilic cytoplasm<sup>1,2</sup>. In our case, the histopathological findings were in accordance with the above mentioned features of angioleiomyoma.

The histopathological diagnosis of leiomyoma is relatively difficult because of the resemblance to many other tumors with spindle-shaped cells. The differential diagnosis would include myofibroma, hemangiopericytoma,

neurofibroma, neurilemmoma, nodular fasciitis, fibrous histiocytoma and schwannoma<sup>3,4</sup>.

Immunohistochemically, leiomyomas are reactive with vimentin, desmin,  $\alpha$  smooth muscle actin and muscle specific actin. In our case immunohistochemical findings revealed positivity for vimentin, smooth muscle actin and negative for S – 100 protein. These features helped us to exclude fibroblastic and neural tumours while indicating myogenic differentiation of the tumour.

The malignant transformation rate has not been reported in the literature<sup>2</sup>. In general, from clinicoradiographic data the tumour doubling time and the resorption of surrounding areas are suggestive of malignancy. From the histologic point of view, the mitotic count has been considered as the most important parameter for differentiating benign and malignant smooth muscle tumours. In our case, though there was rapid increase in size, the tumour had minimal mitotic count which suggests that our case is benign. Despite these findings, it must be remembered that some benign lesions have been metastasized during the later life<sup>1</sup>. The recurrence rate is very low<sup>2, 12</sup>. The line of treatment is conservative surgical excision<sup>1, 2, 6</sup>.

## **Conclusion**

In summary, leiomyoma in the oral cavity is rare and in mandible is still a rarer entity due to paucity of the smooth muscles. But it should be considered in the clinical differential diagnosis in the paediatric patients due to its wide age range of occurrence. Even though in the present case, clinicoradiographic and histologic findings favoured a benign process but a careful periodic evaluation should be planned as benign lesions have metastasized after latency period.

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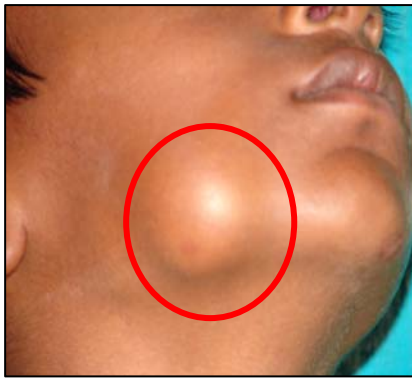


Fig-1, extraoral swelling on right body of the mandible



Fig-2, Panoramic radiograph revealed faint periosteal reaction in the vicinity of permanent first molar and developing tooth buds of right canine and premolars



Fig-3, The axial CT view in bone window showed a massive periosteal reaction on the buccal surface demonstrating the sclerotic borders of the lesion which soft tissue density within the central zone.

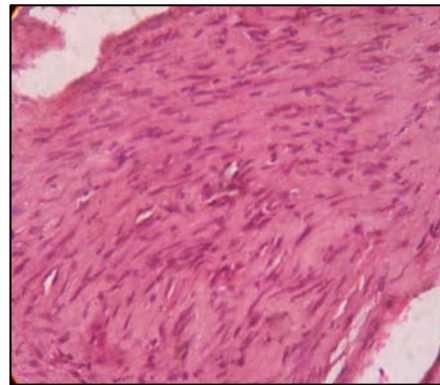


Fig-4, 10x, spindle cells arranged in interlaced fascicular pattern, showing elongated nuclei with blunt ends

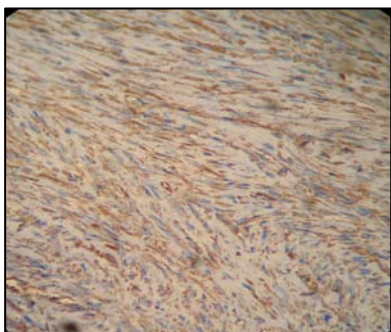


Fig-5, The tumour was positive for vimentin

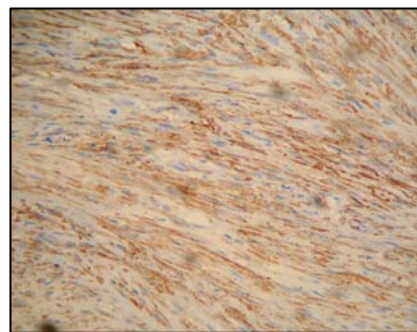


Fig-6, The tumour was positive for  $\alpha$ -smooth muscle actin

COVER LETTER

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
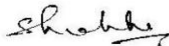



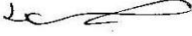
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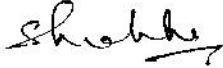



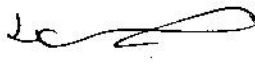
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