

# Unexpected Diagnosis of a Solitary Fibrous Tumour Originating from the Lingual Alveolar Mucosa – A Case Report

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## ABSTRACT

**Introduction:** Extra pleural Solitary fibrous tumours are rare. In the oral cavity they are mainly seen in the buccal mucosa. Clinical diagnosis is often challenging, as they resemble other neoplasms in the oral cavity with no specific clinical features. Usually the diagnosis is made from the histopathological features, following the excision of the lesion and confirmed by immunohistochemistry analysis. As histopathological features are not specific for SFTs immunohistochemistry plays a major role in the final diagnosis of this tumour.

**Case Presentation:** We present a case report on a 36-year-old male patient who presented with an asymptomatic submucosal lump originating from the lingual alveolar mucosa on the anterior mandible, which is a rare site for SFTs in the oral cavity. Diagnosis of this lesion was confirmed following the excisional biopsy which showed features of SFT in the histology with strong immunohistochemistry positivity for CD34, Vimentin and STAT6 (nuclear).

**Key words:** Solitary fibrous tumour, alveolar mucosa, immunohistochemistry, neoplasm.

## INTRODUCTION

Solitary Fibrous Tumour (SFT) is a rare soft tissue tumour that originates from the mesenchymal tissues<sup>1</sup>. In the WHO 2020 classification of soft tissue tumours, Solitary fibrous tumour has been classified under Fibroblastic/myofibroblastic tumours and further sub classified into Intermediate (Rarely metastasizing) and malignant tumour types<sup>2</sup>. SFTs are mainly seen in the pleura and very rarely seen in other sites<sup>3</sup>. In the head and neck region, it comprises 6% of all SFTs and majority of them are seen in the oral cavity<sup>4</sup>. SFTs in the palate, buccal mucosa and floor of the mouth are reported in the literature<sup>3,5</sup>.

Our case report is on an unexpected diagnosis of a solitary fibrous tumour originating from the alveolar mucosa where SFTs are not commonly seen.

## CASE REPORT

36 year old male patient was referred with the history of asymptomatic slow growing lump on the lingual gingivae of the lower premolar region with the duration of six months. The past medical history was unremarkable.

Clinical examination revealed semisolid 1.5cm diameter smooth sessile lump originating from the lingual alveolar mucosa of the lower right premolar region (Figure 1). Orthopantomogram did not reveal any bony involvement.

The patient was scheduled to have a diagnostic biopsy of the lesion under local anaesthesia and during the procedure as the lesion was very fragile a local excision of the lesion was carried out.

Histopathology was reported as moderately cellular tumour which was formed of spindle-shaped cells arranged

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haphazardly and in short fascicles with plump to elongated fusiform nuclei with a dispersed chromatin pattern and inconspicuous nucleoli. (Figure 2,3,4)

By immunohistochemistry of the lesional spindle cells were diffusely and strongly positive for CD34, ( figure 5,6) vimentin and STAT6 (nuclear) (Figure 7,8). The morphological and immuno histochemical features were consistent with SFT.

Complete healing of the surgical site was noted on review. As the tumour margin abuts the periphery and due to the risk of local recurrence the patient has been followed up in the unit. During the first year no local recurrence has been noted.

## DISCUSSION

SFTs in the extra pleural sites compromise 30% - 40% of the SFTs and have been reported to occur in the orbits, liver, extremities, oral cavity, thyroid glands and various other



**Fig. 1:** Smooth sessile lesion originating from lingual alveolar mucosa.

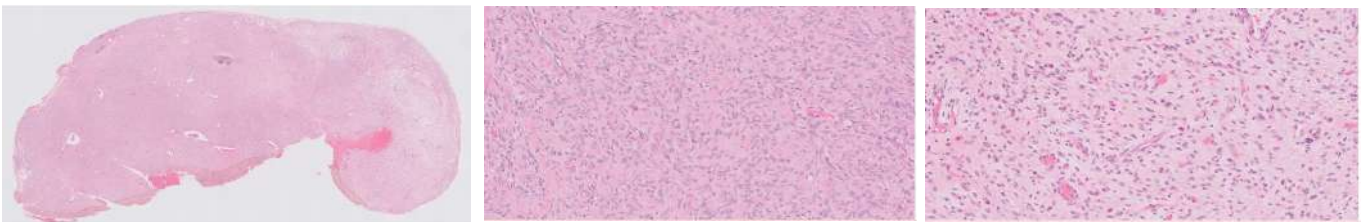
organs<sup>6</sup>. In the oral cavity, buccal mucosa is the most common site followed by tongue. ‘O’Regan et al’ described 21 SFTs in the oral cavity originating from buccal mucosa, tongue, lip, vestibule, pharynx temporal fossa and alveolar mucosa. Among the 21 cases only two cases have originated from the alveolar mucosa<sup>7</sup>. In a study by T Shmuly et al on SFTs diagnosed in a 10 year period, in a single centre 17 were in the head and neck region and 7 were in the oral cavity. None of the 7 cases originated from the alveolar mucosa<sup>8</sup>.

There is no gender predilection noted on a study conducted by J. Gold et al on 79 patients presented with SFT in all anatomical sites. Age range of the patients was 26-82 years<sup>10</sup>.

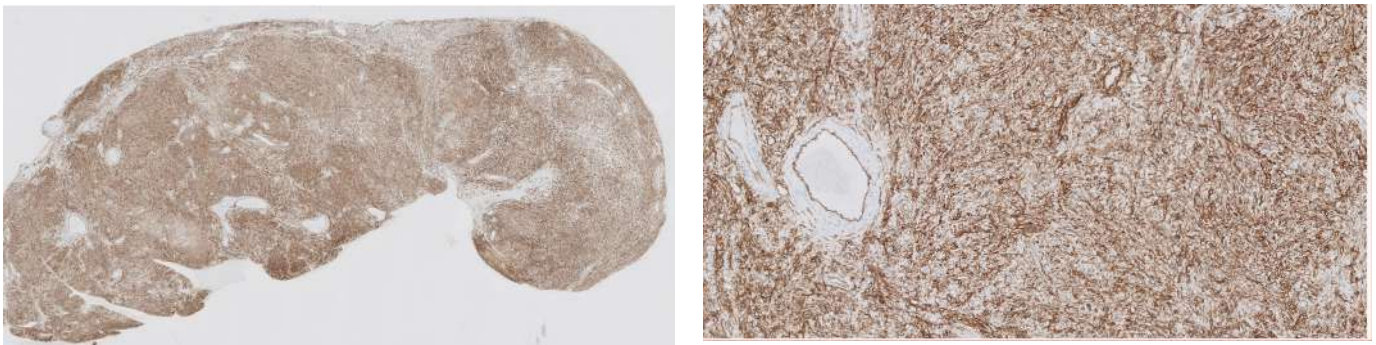
SFT in the oral cavity presents as an asymptomatic submucosal well circumscribed semisolid lump with smooth overlying mucosa as in our case<sup>9</sup>. Clinically they could mimic a minor salivary gland tumour<sup>3</sup>. We included minor salivary gland tumour in our differential diagnosis.

Investigations recommended for SFTs include, Biopsy and FNA. MRI or CT scans are not routinely requested for intra oral SFTs unless bony involvement is suspected.

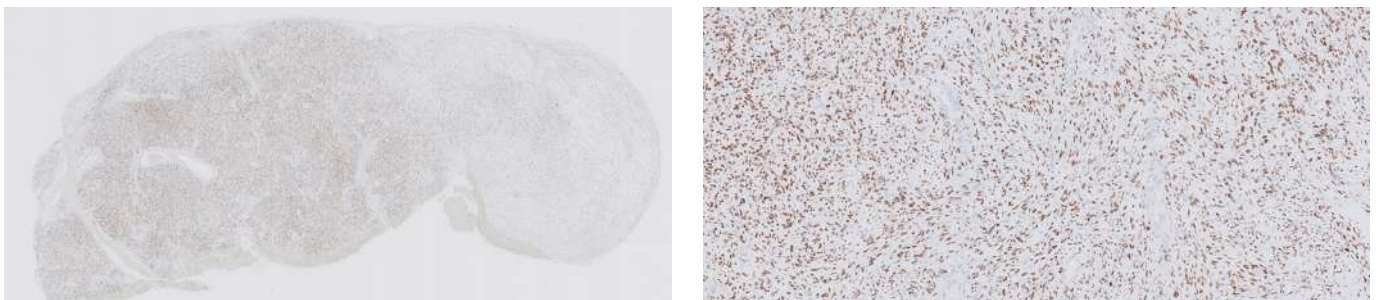
SFTs are composed of spindle shaped cells with little



**Figs. 2,3,4:** Spindle-shaped cells arranged haphazardly and in short fascicles with plump to elongated fusiform nuclei with a dispersed chromatin pattern and inconspicuous nucleoli.



**Figs. 5,6:** Lesional spindle cells were diffusely and strongly positive for CD34



**Figs 7,8:** Lesional spindle cells were diffusely and strongly positive for STAT6

cytoplasm in collagenous background. There is no obvious pattern in which the spindle cells are arranged but densely cellular and hypocellular areas are common histological findings in SFTs. Prominent vascularity with haemangiopericytoma is also a feature of SFTs<sup>3,5,11</sup>.

Chan et al<sup>12</sup> stated that immunohistochemical study is essential to confirm the diagnosis of extrapleural SFTs. Several studies have confirmed strong immunohistochemical expression of CD34 in SFTs<sup>7-14</sup>. Positivity for Vimentin, bcl-2, CD99 are also reported as additional markers<sup>13,14,15</sup>. In our case nuclear expression of STAT6 was strongly positive. Leona A doyle et in their study, stated that STAT6 is highly sensitive and almost a specific immunochemical marker for SFTs and recommended STAT6 as a diagnostic marker for SFTs<sup>16</sup>.

Local recurrence is rare in oral SFTs even on tumours excised with involved margins<sup>7,8</sup>. On an study by O'Regan et al on 21 patients, margins were positive in 18 cases with no history of recurrence during the period they were reviewed<sup>7</sup>. Incomplete resection of the SFTs in the head and neck region is not uncommon due to the complex head and neck anatomy.

Due to the rarity of the tumour and limited number of studies on SFTs in the oral cavity, there is only very little data available on prognosis and late recurrence on tumours existed with involved margins.

## CONCLUSION

SFT in the oral cavity is rare and SFT originating from alveolar mucosa is even more rare. As the clinical features resemble the other neoplasms, it can be challenging to diagnose from the clinical features itself. SFTs should be considered in the differential diagnosis of submucosal lumps in the oral cavity. They are usually diagnosed following the excision of the lesion. Bi immunohistochemistry is widely used to confirm the diagnosis. Complete excision may not be achievable in most cases in the oral cavity, without disfigurement due to the complex anatomy. Recurrence is not reported even on tumours resected with positive margin after long term follow up .

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