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Case Report

Solitary fibrous tumor with high proliferative index arising from floor of the mouth

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1. Introduction

Solitary fibrous tumor (SFT) is a rare entity in the maxillofacial area that resemble with spindle cell tumor. The very first case of SFT (arising from pleura) was reported by Klemperer and Rabin in 1931 [1]. Recent studies revealed that SFT is mesenchymal in origin and they are also reported in extra-serosal sites [2,3].

We report an interesting case of SFT originating in the floor of the mouth with high proliferative index.

2. Case report

A 48 years old male presented with a 3 month history of slowly enlarging, painless swelling in the right lingual vestibule causing dysarthria and difficulty in mastication. On clinical examination, the lesion was well circumscribed, 5 cm in size, non-tender, firm in consistency, raised floor of the mouth with normal overlying mucosa and tongue displaced on right side (Fig. 1). No facial asymmetry seen on extra-oral examination. Patient has no history of trauma and surgery of the floor of the mouth. Contrast-enhanced computed tomography (CT) scan revealed a large homogenous mass which was displacing the sublingual gland laterally with no evidence of bony involvement (Fig. 2). The tumor was excised completely under general anesthesia through submandibular approach (Fig. 3). The specimen was sent for histopathological and immunohistochemical examination.

Gross examination revealed an encapsulated ovoid, solid, grey-white tumor with a smooth capsular surface measuring 5.5 × 4.5 × 3 cm in size. No areas of necrosis or hemorrhage were seen. Histopathological examination revealed a tumor composed of spindle shaped cells disposed in form of intersecting long and short fascicles (Figs. 4 and 5). The tumor was well-vascularised and showed presence of staghorn blood vessels. Tumor showed foci of increased cellularity with focal moderate to marked nuclear pleomorphism and increased mitoses (Fig. 6). Mitotic index was 5–7/10 hpf (high power fields). There were no areas of necrosis. Tumor was limited by capsule. No infiltrative borders were seen. Immunohistochemically, the tumor was positive for CD34 (Fig. 7) CD99, Bcl-2 (Fig. 8) and vimentin (Fig. 9) whereas negative for Pan CK, EMA, S-100, SMA, CD 117 and CD31. Ki-67 (proliferation index) was found to be 25%.

On the basis of histopathology and immunochemistry a final diagnosis of Solitary fibrous tumor with high proliferation index was made. Patient is currently under observation and he is free of any recurrence/disease at the operated site after a 2 year follow up.

3. Discussion

Solitary fibrous tumor (SFT) is rare spindle cell neoplasm mostly arising from pleura or serosa. Initially it was considered to be of mesothelial origin. Few SFTs has been reported in various extra-serosal sites as well which favour its mesenchymal origin and is not confined to serosa [2–4]. SFT developing at extra-pleural sites creates some difficulty in its diagnosis as they are very rare and poses similarity with other neoplasms e.g. sarcoma [5,6]. SFT arising from floor of the mouth or salivary glands can be differentially diagnosed as salivary gland tumors, ranula, schwannoma, histiocytoma or haemangiopericytoma [6–8].

According to Shine et al. [8] 40 cases of SFT in oral cavity were reported involving tongue, buccal mucosa, lip, gingival, sublingual glands, soft and hard palate, mental region. However, they have not mentioned the origin of SFT from non-salivatory soft tissue of floor of the mouth. Cox [9] reported 153 cases of SFT in the head and neck region and observed that buccal mucosa is frequently involved as compared to nasal cavity, pharyngeal area and tongue. Many authors reported cases of SFT of the floor of the mouth.

O'Regan et al. [3] reviewed 21 cases of solitary fibrous tumor presenting in the oral cavity and described their clinical, morphologic, and immunohistochemical features in detail.Clinically, SFT of oral cavity are well circumscribed, non-tender, slow growing mass firm in consistency. Occasionally a large size lesion can cause difficulty in mastication and dysarthria [8,10–13].

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Fig. 1. Well defined swelling in the floor of mouth extending from 35 to 48 region.

Kurihara [2], O'Regan et al. [3], Ferreiro and Nascimento [6], Ogawa et al. [7], Sousa et al. [10], Carlos et al. [11], Rodrigues et al. [13] described similar microscopic and immunohistochemical features for SFT. They reported a slowly enlarging mass, solid, firm in consistency, grayish-white in color. Microscopic examination revealed well demarcated, spindle cells in collagenous fibrous tissue in "Staghorn configuration", alternating Hypocellular and hypercellular areas, oval nuclei, scant eosinophilic cytoplasm, with abundant hyalinsed blood vessels.

Immunohistochemically, the tumor was positive for CD-34, CD-99, Bcl-2 and vimentin; whereas negative for desmin, actin, S-100 and CD68. These findings were similar to the diagnostic criteria for SFT proposed by Chan [14]. Microscopically, malignant SFTs are usually hypercellular lesions, showing at least focally moderate to marked cytological atypia, tumor necrosis, numerous mitoses (over 4 mitoses per 10 HPF) and/or infiltrative margins. In present case, tumor showed focal moderate nuclear atypia, increased cellularity, mitotic index > 4/10 hpf and higher Ki-67 (Ki-67- 25%); however marked nuclear atypia, tumor necrosis and infiltrative margins were not seen.

Ki-67, a proliferation-associated antigen, is expressed in active phases of the cell cycle including G1, S, G2, and mitosis. Sun et al. [15] performed immunohistochemical staining for Ki-67 in 24 cases of benign (14 patients) and malignant (10 patients) SFT. Their results showed that the mean Ki-67 labeling index is 1.9% for benign SFTs and 6.11% for malignant SFTs ($P < 0.05$). It suggested that Ki-67 could be

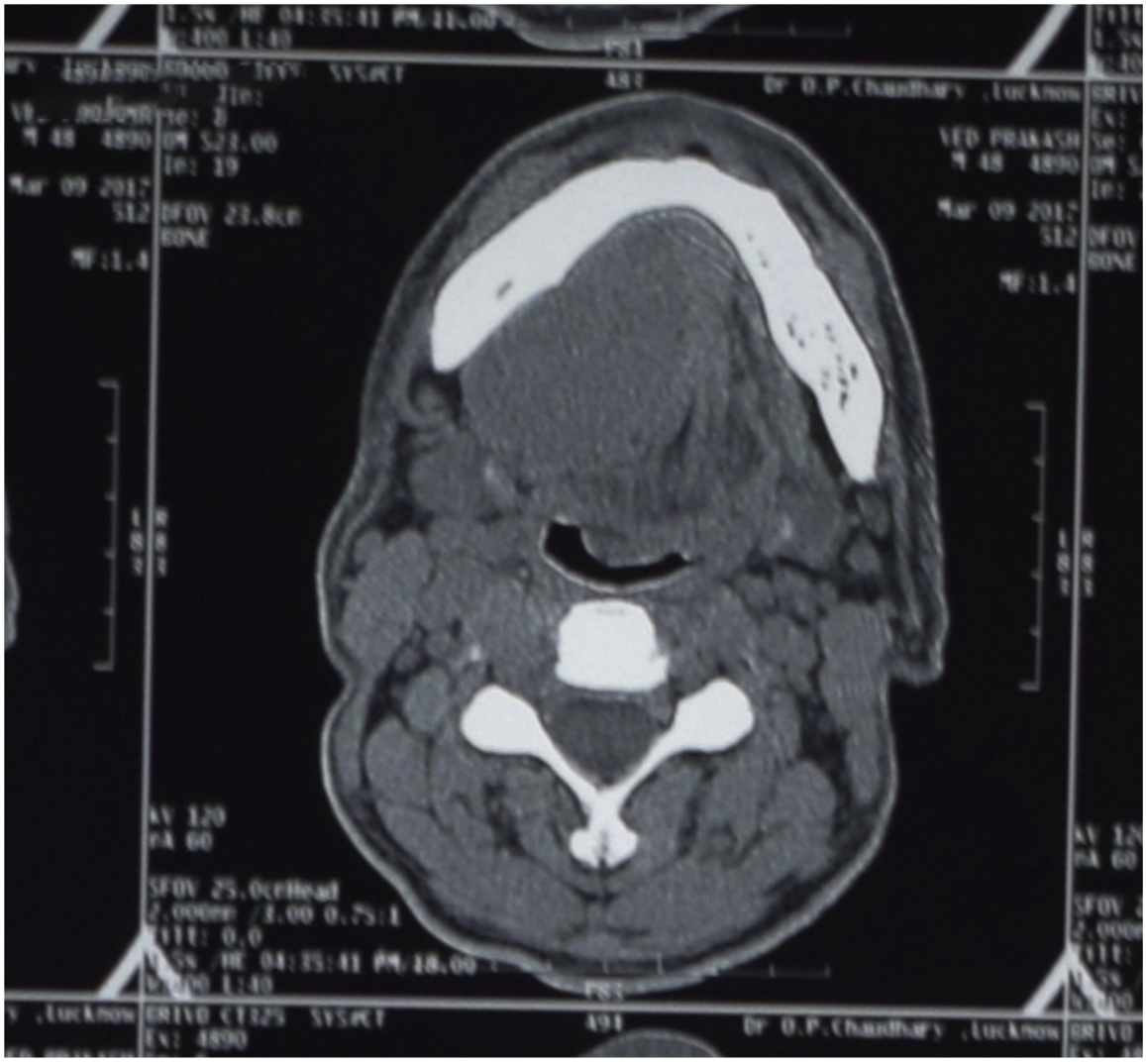

Fig. 2. Axial CT of the floor of mouth showing on right side, a well circumscribed ovoid lesion, displacing submandibular gland laterally.

diagnostically relevant to the evaluation of malignant SFT [9]. Present

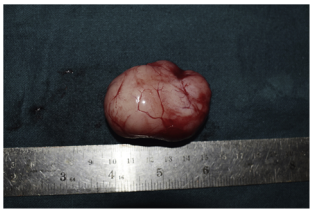

Fig. 3. Excised specimen.

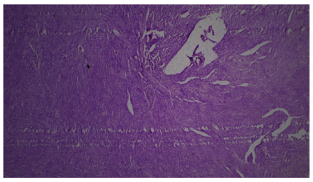

Fig. 4. Haematoxylin and eosin stain in ×20 magnification showing a proliferation of spindle shaped cells arranged in fascicles.

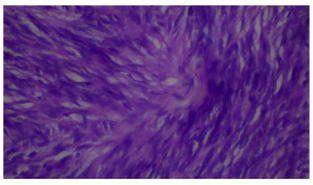

Fig. 5. Haematoxylin and eosin stain in ×40 magnification showing Collagenous bundle with spindle shaped tumor cells.

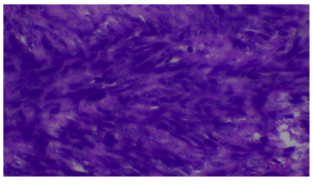

Fig. 6. Haematoxylin and eosin stain in ×40 magnification showing spindle shaped tumor cells.

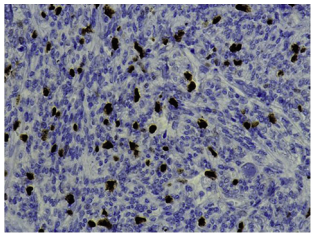

Fig. 7. Tumor immuno-histochemically positive for CD-34.

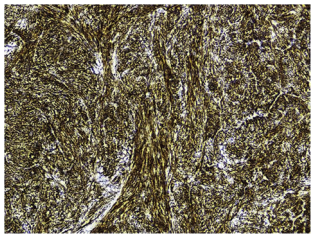

Fig. 8. Tumor immuno-histochemically positive for bcl-2.

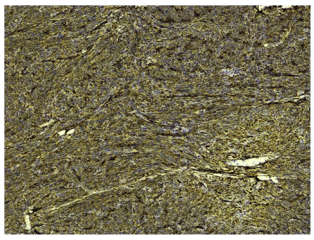

Fig. 9. Tumor immuno-histochemically positive for vimentin.

case showed high Ki-67 which highlighted that tumor is capable of high degree of proliferation; however all criteria for malignant SFT's were not met. Hence patient was put on clinical long term follow-up.

SFT at non-pleural/extra-serosal sites are usually benign [16–18] and local excision of the tumor is the treatment of choice [12,18,19]. The adjuvant therapies such as chemotherapy and radiotherapy have been reported where surgical excision was not possible [9,20]. Surgical excision is the treatment of choice for both benign and malignant SFTs. In our case the tumor was completely resected and no adjuvant treatment was given. Patient is on regular follow-up and has shown no signs of recurrence.

EF de Morais et al. [12] have reviewed characteristics of six reported cases of solitary fibrous tumors in floor of mouth. All cases presented as well-defined nodules with average size of 4 cm in largest dimension and none showed microscopic features of malignancy. Our case is the tenth case described in literature at this site [7,8,11–13,19,21–23] and differs from the earlier described cases by the fact that it showed high proliferation index and increased mitoses (Table 1).

The diagnosis of SFT affecting extra-pleural sites is difficult because of the nonspecific clinical and microscopic features of the tumor. Overall, SFTs of the head and neck have a substantial local recurrence rate; indeed exceeding that seen for SFTs at other sites, but only infrequent ones metastasizes or cause death from disease. [24] We agree with the suggestion by Smith et al. [24] that the high rate of local recurrence underscores the intermediate malignant potential of these tumors and the need for long term follow-up.

SFT of the floor of mouth is rare and difficult to diagnosis but thorough analysis of clinical, histopathological and immunohistochemical features helps to establish the diagnosis of the tumor. SFT should be included in the differential diagnosis of tumors arising from soft tissues of the floor of the mouth. Suitable treatment and long term follow up is important for good prognosis.

Ethical approval

Ethical clearance was obtained from research and ethical committee of Sardar Patel Postgraduate Institute of Dental And Medical Sciences, Lucknow.

Table 1
Cases of solitary fibrous tumor located in the floor of the mouth.

S.No.	Report (name, reference)	Year	Age (years)	Sex	Size (mm)	Follow-up (months)	Recurrence
1.	Ogawa et al. [7]	2003	59	M	38 × 30	27	No
2.	Shine et al. [8]	2006	35	F	30 × 40	5	No
3.	Ayad and Ghannoum [21]	2007	74	F	30		No
4.	Tsuji et al. [22]	2008	84	M	15 × 12 × 9	12	No
5.	Nobusawa et al. [23]	2015	88	M	50 × 45 × 20	6	No
6.	Shi and Wei [19]	2015	39	F	30 × 40	22	No
7.	Carlos et al. [11]	2016	70	F	40	24	No
8.	Morais et al. [12]	2017	66	F	40	7	No
9.	Rodrigues et al. [13]	2017	54	M	28 × 23 × 14	96	No
10.	Present case	2018	48	M	55 × 45	24	No

Conflict of interest

The authors declare that they have no conflicts of interest.

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