



Solitary fibrous tumour of the parotid gland: a case report and a 15-year literature review

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Abstract: Solitary fibrous tumour (SFT) is uncommonly found in the salivary glands and is a rare group of spindle-cell tumours. A review of literature revealed only 40 reported cases of SFT of major salivary glands over a 15-year period from 2004 to 2018. SFTs of the salivary glands are usually benign, although rarely, can be aggressive and may sometimes recur after initial resection. Histology and Immunohistochemistry are the most important criteria to distinguish SFT from other head and neck tumours. SFTs strongly stain for CD34 and hence is the most frequently used stain for diagnosis. All reported SFT cases with available information on immunohistochemical stains were positive for CD34. Recently, immunohistochemistry for STAT6 has been introduced as a surrogate diagnostic marker for SFT that is highly sensitive and specific. We report a case of a 51-year-old Chinese gentleman who presents with a 3-month history of a left parotid tumour that required a subtotal parotidectomy for complete excision. It was diagnosed as SFT based on histology and immunohistochemical features. He was followed-up for 3 months post-operatively with no clinical evidence of recurrence. This case proposes that, although rare, SFT should be considered in the differential diagnosis of soft-tissue tumors in the major salivary glands. An awareness of this rare entity will help clinicians and pathologists better manage similar patients in the future.

Keywords: Solitary fibrous tumour (SFT); hemangiopericytoma; major salivary gland; parotid

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Introduction

First discovered in the pleura, solitary fibrous tumour (SFT) was previously falsely thought to be of mesothelial origin. It is then shown to be a fibroblastic mesenchymal neoplasm likely from adult mesenchymal stem cells hence giving rise to a variety of anatomical origins (1). SFTs in the head and neck region comprise about 6% of all SFTs in previously published data (2). Of the head and neck SFTs, the most common sites involved are the sinonasal tract and orbit, followed by oral cavity, then salivary glands (2). A review of literature found 40 reported cases of SFT in major salivary glands from 2004 to 2018. Here we report an additional case of SFT arising from the left parotid gland as well as a 15-year review of the literature on SFTs arising from major salivary glands.

Case presentation

A 51-year-old Chinese gentleman presented with a 3-month history of a left angle of mandible lump. Physical examination showed a left angle of mandible 2.5 cm firm mass that was non-tender, with no overlying skin changes. The facial nerve function was normal, and there were no other palpable lumps on examination of his neck.

Preoperative computed tomography (CT) scan of the neck showed a 4.2 cm × 2.9 cm × 3.3 cm fairly well margined homogeneously enhancing lesion within the left parotid gland (*Figure 1*). There was no central necrosis or adjacent stranding. Fine needle aspiration cytology showed cells which appear basaloid, with minimal stroma suggestive of an aspirate from a basaloid lesion, including basal cell adenoma and a cellular pleomorphic salivary adenoma.



Figure 1 Axial CT scan after contrast injection showing a homogeneously enhancing 4.2 cm × 2.9 cm × 3.3 cm mass within the left parotid gland.

The patient underwent a left subtotal parotidectomy. Post-operatively, the patient recovered uneventfully, and has recovered well from a partial left facial nerve paresis.

The excised specimen was a 6 cm lobulated fleshy tumour involving both superficial and deep lobes of the parotid gland, with epicentre likely in the deep lobe. The tumour lies lateral to the retromandibular vein but deep to the facial nerve.

Figure 2 show the microscopic appearance of the tumour. The tumour is cellular with short fascicles of plump spindle cells with little intervening fibrous stroma, displaying a hemangiopericytomas pattern. There are no areas of de-differentiation. The margins were not infiltrated by tumour, although very close, focally <0.1 mm. The high power view show spindle cells displaying oval nuclei without any significant atypia. Immunohistochemical studies show that the neoplastic cells diffusely express CD34, bcl2 and CD99, and nuclear expression of STAT6. There was no expression of cytokeratins (AE1-3), CK14, SMA, CD21, CD35 or p63. The Ki-67 proliferation index was approximately 10–20%. Hence, a diagnosis of SFT was made.

The patient was discussed at a multi-disciplinary head and neck tumour board and decision was made to offer post-operative Radiotherapy. His tumour margins were considered to be close, and Ki 67 proliferation index was not low. He was followed-up for 3 months with no clinical evidence of recurrence. However, the patient declined adjuvant treatment.

Discussion

Most SFTs of the parotid origin reported in the literature are of the fibrous variant characterised by alternating areas of fibrous hypocellularity and hypercellularity composed of round-to-spindle cells, tapering cytoplasm with a fascicular, storiform arrangement (3,4). They are associated with “staghorn” vasculature.

It is also thought to be synonymous with Giant Cell Angiofibroma and Hemangiopericytomas. Since 2002, the terms Hemangiopericytoma and SFT had been unified under the term SFT according to WHO classification of soft tissue and bone tumors, and in 2013 the term Hemangiopericytoma has been replaced by SFT (4).

SFT of the major salivary gland are a rare group of spindle-cell tumours. We did a literature review in PubMed using the search terms “Hemangiopericytoma” and “Solitary Fibrous Tumour” in combination with “Sublingual Gland”, “Submandibular Gland”, “Parotid Gland” “Salivary Gland”. We selected articles published within the time period of January 2004 to October 2018. Clinical series of head and neck SFTs were included if critical information on salivary gland SFTs were reported. Foreign language articles were included only if an English version was available, and duplicated cases reported in multiple articles were excluded.

Including our case reported here, only 40 cases have been reported over the past 15 years from 2004 to 2018. Thirty-four of which were from the parotid gland (including our case), five were from submandibular gland and one from the sublingual gland. *Table 1* summarises the characteristics of these reviewed cases (1,2,5–26).

Clinical presentation and investigations

Majority of reported cases involve the parotid gland, rather than the submandibular and sublingual glands. The age range of patients with major salivary gland SFTs is very wide from 11 to 79 years (mean 48.52 years). Although SFTs are not known to have any gender predilection, our review showed a slightly higher incidence of SFTs in men (54%) as compared to women (46%). This is similar to findings in other studies done on head and neck SFTs (8).

SFT of the parotid and other major salivary glands usually present as a slow growing, palpable non-tender mass. Occasionally, they may cause compression

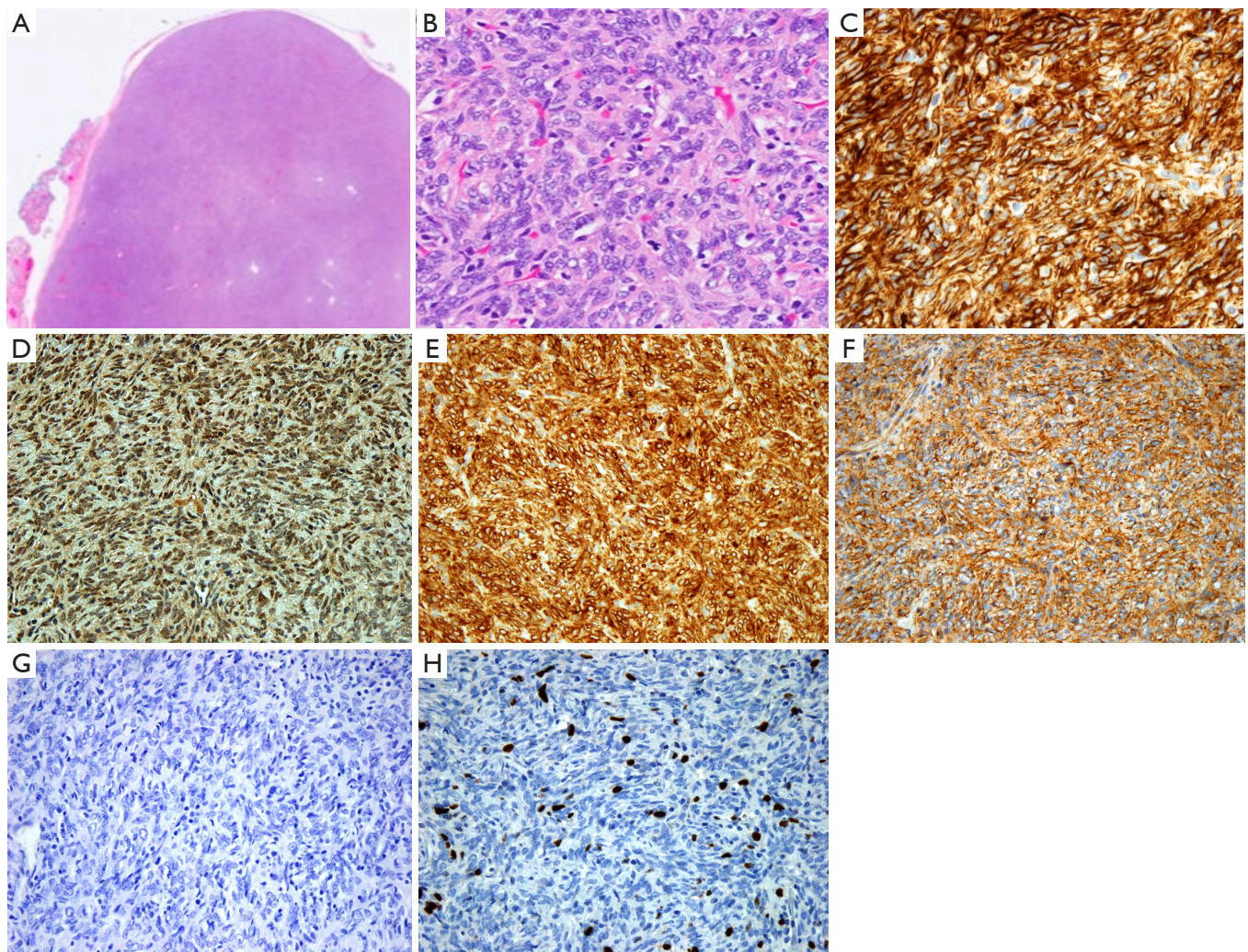


Figure 2 Microscopic appearance of patient's tumour with corresponding stainings. (A) H&E stain low power $\times 20$ magnification view showing circumscribed borders; (B) high power view H&E stain $\times 400$ magnification; (C,D) figures showing positivity to CD34 and STAT6 respectively $\times 400$ magnification; (E,F) figures showing positivity to Bcl 2 and CD99 respectively with $\times 400$ magnification; (G) $\times 400$ magnification showing negative staining for smooth muscle actin; (H) $\times 400$ magnification showing Ki67 proliferation index 10–20%.

symptoms such as dysphagia, in which case extension to the parapharyngeal space is suspected (27). Clinical presentation is similar to that of other tumors such as pleomorphic adenomas and lipomas. Patients complain of a firm and well-defined mass that is slow-growing over a period of months to years (*Table 1*). FNAC is rarely helpful. Radiographic findings are also nonspecific. Magnetic resonance imaging usually shows an isointense mass on T1-weighted images and high signal intensity with enhancement in T2-weighted images (18). Therefore, a diagnosis of SFT is made frequently based on histological and immunohistochemical analysis.

Histology and immunochemistry

Macroscopically, SFTs are known to be well-circumscribed, firm, white-tan or gray and encapsulated.

CD34 has been shown to be a very useful marker in the diagnosis of SFT. Only a portion of the cases found reported Immunohistochemical results, and the stains performed vary across literature. We subanalysed cases that had reported stainings for the three most common markers namely CD34, bcl-2 and CD99, summarised in *Table 2*.

This high rate of positivity is consistent with studies done in SFTs of other anatomical locations (5,28,29) Bcl-

Table 1 Summary of reviewed cases in English language literature from 2004 to 2018

| Characteristic | Number (n=40) |
|--------------------------------|---------------|
| Anatomical site | |
| Parotid | 34 |
| Submandibular | 5 |
| Sublingual | 1 |
| Gender | |
| Males | 22 |
| Females | 18 |
| Ratio | 1.17:1 |
| Age (in years) | |
| Range | 11 to 79 |
| Mean | 48.52±17.29 |
| Median | 46.50 |
| Size (cm)* | |
| Range | 1.0–6.5 |
| Mean | 3.66±1.85 |
| Median | 3.45 |
| Duration of symptoms (months)* | |
| Range | 0.25–120 |
| Mean | 27.65 |
| Median | 12 |
| Mode | 12 |

*, Not reported for some cases.

Table 2 Summary of major salivary gland SFT cases with reported stainings for CD34, bcl-2 and CD99

| Reported stainings from major salivary gland SFT cases | CD34 | bcl-2 | CD99 |
|--------------------------------------------------------|----------|----------|----------|
| No. of cases reported to be positive | 21 of 21 | 13 of 16 | 13 of 15 |
| Percentage % | 100 | 81.3 | 86.7 |

2 may be positive in other kinds of mesenchymal tumors like sarcomas, hence are not as specific for SFT (8). In some cases of malignant and dedifferentiated SFT, CD34 may be negative. Hence, CD34 is useful in combination with Bcl-2 as a consistent characteristic of SFT (29).

Immunoreactivity to STAT6 has been shown to be highly sensitive and specific for SFTs (28). This finding is useful, as less than 10% of other spindle cell tumors are positive for STAT6 (29). In our case, STAT6 was used as an adjunct to diagnosis and found to be positive. Although none of the other salivary gland SFTs in our review were tested for STAT6, a multi-institutional clinicopathological study on SFT in other anatomical sites found that all 45 out of 45 (100%) cases showed positivity to STAT6 (28). Yoshida *et al.* also published a validation study where also 49 out of 49 (100%) SFTs showed STAT6 expression, and only a 2.5% (4 out of 155) of cases of other tumor mimics of SFT showed weak nuclear STAT6 expression (30).

Treatment and outcome

Complete surgical resection is the prevailing treatment, and most authors recommend long-term follow-up for all patients with SFT due to the possibility of recurrence and a minority behaving aggressively (31). The pathological features used to predict poorer outcomes in SFT tumors of the head and neck region are hypercellularity, nuclear atypia, more than 5 mitoses per 10 high-power fields and tumour necrosis (2,4). These features may not be the sole predictor of behaviour as some SFTs that do not meet the above criteria are shown to have recurred. Although reported in a minority of SFT involving other anatomical sites, metastases have not yet to be reported in SFTs of the head and neck region (31).

There were two cases of Parotid SFTs in 2014 and 2015 that reported recurrence at 24 and 20 months post-surgery respectively (6,8). Involved margins was only reported in the latter case. Ki 67 proliferation rate of <4% is generally considered benign, and >20% considers the tumour a malignant variant (8,32). However, in these two cases, Ki67 rate was reported as 5% and 5–10% respectively, a range that is considered intermediate. In both cases, adjuvant radiotherapy was not administered.

Ganly *et al.* (33) suggest that patients with positive surgical margins or whose tumors have a malignant component benefit from adjuvant postoperative radiotherapy. However, there are few reports concerning adjuvant treatment and their follow up periods are of short duration. Longer term studies in this area may be beneficial.

Conclusions

In summary, this case study reports a rare case of solitary

fibrous tumour in the parotid gland occurring in a 51-year-old Chinese gentleman. Diagnosis of this tumour was made mainly based on histology and immunohistochemical staining namely CD34 used in combination with Bcl-2 and STAT6. Its presentation, epidemiology and size is consistent with the ranges reported in literature of other SFTs of the major salivary glands over the past 15 years. Complete surgical excision is the most widely accepted treatment, with a lack of information surrounding the efficacy of radiotherapy. Hence, SFTs occurring in the major salivary glands should be followed-up with care considering their unpredictable course and potential for recurrence.

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None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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