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Solitary Fibrous Tumour (SFT) of Parotid Gland : A case report

Bandana Mehrotra[(1)], Sanjay Mehrotra[(2)], Shubhi Pandey Sharma[(3)],Ashok Kumar Kapoor[(4)], Mithlesh Bhargava [(5)], Supriya Mehrotra[(6)], Saumya Brij[(7)]

(1) Director & Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(2) Director, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(3) Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(4) Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(5) Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(6) Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.
(7) Pathologist, Department of Pathology, RML Mehrotra Pathology Pvt. Ltd,, Nirala Nagar, Lucknow, Uttar Pradesh, India.

Abstract
Introduction: *SFT is a rare spindle cell tumor that is usually associated with pleural or peritoneal neoplasms. SFT of the parotid gland is a rare occurrence, only 38 cases of it including our case have been reported in the literature all over the world. Parotid gland solitary fibrous tumour (PG-SFT) is a well-circumscribed encapsulated tumour with interlacing bundles of the spindle to epithelioid cells. Tumour is hypercellular and composed of cells having round to oval nuclei with coarse chromatin. Its histogenesis is not known. Probably, it is derived from adult mesenchymal stem cells. Tumour is highly vascular. Tumour cells are positive for CD 34, Bcl2, and CD99 immune markers and negative for S100, cytokeratin, EMA, and smooth muscle actin. D/D includes pleomorphic adenoma, myoepithelioma, schwannoma, and spindle cell melanoma. Herewith, we describe the features of a case of solitary fibrous tumour (SFT) of the parotid gland.*
Case report: *A 32 year old female had a slowly increasing tumour of left parotid gland. Tumour was round 4×3x×2 cm. Patient noticed tumour since 2 years. Tumour gradually increased in size. There was no redness. Tumour was well- circumscribed. Surgical excision of the tumour was done. Grossly, several pieces of tumour tissue were received for histopathological examination. Microscopic examination showed hypercellular tumour. It consisted of spindle cells with streaks of collagen. Tumour cells had central vesicular nuclei with eosinophilic cytoplasm. A draining lymph node with reactive lymphocytic hyperplasia was also seen.*
Discussion : *A middle aged man had a benign solitary fibrous tumour in parotid gland. It measured 4×3×2 cm. Tumour was excised. Microscopically, tumour consisted of alternate hypercellular and hypocellular fibrous areas of spindle cells. Ki-67 proliferative index was 4% suggesting its benign nature. Tumour was completely excised and the patient appeared cured.*
Conclusion: *Initially, the current tumour was misdiagnosed as a localized mesothelioma. Later, it was identified as a mesenchymal tumour and was renamed as solitary fibrous tumour(SFT). Thus SFT should be considered as a significant differential diagnosis in slow growing well defined solid tumors to avoid aggressive treatment. Complete surgical resection of the tumor is the treatment of choice.*
Keywords: *Hemangiopericytoma, Benign, Recurrence, Malignant potential*

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I. Introduction:

Parotid gland solitary fibrous tumour (PG SFT) is a spindle cell neoplasm and may arise from primitive mesenchymal cells[1]. It is most commonly seen occurring in pleura and peritoneum. Only 37 cases of PG SFT have been reported world-wide till date. These tumours are slightly more common in males (male female ratio being 1.2:1) in all age-groups [2]. Most cases of SFTs are benign. Rarely malignant transformation

noted. First case of PG SFT was reported in 1995 [3]. Tumour may develop as a painless slow-growing benign neoplasm arising from parotid parenchyma. These neoplasms are treated by surgical excision. Infrequently, most of PG-SFT recur. These tumours comprise of spindle cells showing cytoplasmic positive reaction with anti-CD34 and anti-vimentin antibodies. Focal mild reaction with anti-EMA and anti-Bcl2 antibody might be seen. Moreover, Ki-67 proliferative index might vary between 20% to 30%. A single case of PG SFT arising from the Stensen's duct is reported in literature so far.[4]. Also, a single case of dysphagia has been reported due to compression of oesophagus by the tumour[5]. However, most of the cases remain silent and asymptomatic [5].

II. Case Report :

A male patient aged 32 years presented with a swelling of left parotid gland. It was slowly growing painless tumour. On radiological examination, USG findings suggested well defined hypoechoeic lesion with lobulated margins involving deep lobe of parotid gland extending into the superficial lobe. Mild internal vascularity of the tumour along with few subcentimetric intra parotid and periparotid lymph nodes were also noted. Cytological examination findings were suggestive of pleomorphic adenoma. On gross examination, the tumor was well circumscribed encapsulated mass, grayish-white in colour and measured 4×3×2 cm. It was excised and surgically removed. Microscopically, the tumour was hypercellular and composed of interlacing bundles and storiform arrangement of fusiform spindle to epithelioid cells. The cells enclosed oval to elongated nuclei with scanty cytoplasm. Mild nucleomegaly was seen. Irregular thin walled dilated vascular spaces with few staghorn blood vessels were also seen traversing the tumour stroma. Areas of hyalinised and collagenised stroma with scanty mitoses were also noted. No anaplastic areas or areas of necrosis were seen(figure1).

III. Discussion :

SFT-PG is a rare spindle cell neoplasm of mesenchymal origin with only 38 cases of it reported so far in the literature. SFT was first described in 1767, described in detail later by Wagner in 1870. Klemperer and Rabin categorized pleural tumors into diffuse mesotheliomas and localised mesotheliomas also known as solitary fibrous tumour.Initially, the current parotid gland (PG) tumour was misdiagnosed as localized mesothelioma. Later, it was identified as a mesenchymal tumour and was named as parotid gland solitary fibrous tumour (PG-SFT). In addition, most of the previously reported hemangiopericytomas may not be pericytic in origin but instead constitute a cellular variant of SFT[6]. Clinically SFT may mimic a lot of disorders like salivary gland tumours, mesenchymal neoplasms, lymphomas, Sjogren's syndrome, infections, sialadenosis and sarcoidosis. Cytological analysis of SFTs have always had limited significance in the diagnosis of the tumour, with majority diagnoses suggestive of spindle cell neoplasm. Upon radiological examination, all SFTs generally appear non specific. Hence, histology and immunohistochemical study stand as front line investigations for the specific diagnosis of SFT-PG.

Grossly, parotid SFTs are well defined, painless, slow growing, white-tan to grey localised masses with no age or gender predilection. Microscopically, SFTs show alternating hypercellular and hypocellular areas having round to spindled cells arranged in storiform or facicular pattern. Individual cells show round to oval to fusiform centrally placed nuclei and tapering cytoplasm. Stroma may show areas of collagenous matrix with numerous branching and ramifying vessels in a staghorn appearance with thickened or hyalinised walls. IHC is the most important tool for diagnosis of this tumour. In the present case, an initial differential diagnosis of SFT, myoepithelioma and neurofibroma were considered. IHC was performed with STAT6, CD-34, CD-99, BCL2, Ki-67, PCK, GFAP and MYOGENIN. Anti-STAT6 antibody stained nuclei of tumour cells as positive. Tumour cells showed strong diffuse membranous positivity with anti-CD34 antibody. Anti-CD99 antibody showed diffuse strong membranous positivity of the tumour cells. Anti-Bcl2 antibody stained nuclei as well as the cytoplasm of tumour cells. Tumour showed low proliferative activity with anti-Ki67 antibody (<4% of nucleated cells). Anti-PCK antibody failed to stain the tumour cells. Tumour cells showed negative reaction with anti-GFAP antibody. Anti-myogenin antibody showed aberrant positivity of the tumour cells.

Tumours having high cellularity, pleomorphism, necrosis, high mitotic rate and infiltrative margins are likely to behave in an aggressive manner[7-10]. PG-SFT are benign neoplasms with malignant potential[11,12]. It may also present as a malignant neoplasm[2]. In addition, these tumours are well-circumscribed and may be cured after excision[2]. Further, PG-SFT may arise from pleuripotent fibroblasts[13]. These are locally aggressive tumours. These tumours rarely metastasize[2]. Neoplasm of parotid gland may have metastatic potential[5]. Additionally, local invasion of the mandible has been described in PG-SFT[15]. Further, fusion of NAB2 and STAT6 genes has been reported in chromosome 12 in PG-SFT[16]. Furthermore, > 6 mitoses/hpf and a decrease in CD34+ tumour cells may suggest a malignant change[6]. Moreover, >10% Ki-67 positivity may suggest a malignant change while a proliferative index of 4% may suggest a benign neoplasm[17]. Present tumour also had a proliferative index of 4% suggesting a benign neoplasm.

IV. Conclusion :

Although a rare occurrence, SFT should be considered a significant differential diagnosis in parotid gland tumours that are fairly palpable, painless, slow-growing and well circumscribed, to avoid aggressive treatment of the tumour. FNAC examination of such tumours is mostly inconclusive and hence insufficient to reach the final diagnosis. Histopathological examination and IHC, therefore, serve as benchmark investigations for a specific accurate diagnosis.

SFT-PG are benign tumours but may rarely metastasise. Presence of significant cytological atypia, necrosis and frequent mitotic activity are suspicious of malignant transformation. Studies have theorised malignant SFTs show reduced CD34 positivity compared to benign SFTs. Complete local excision with clear surgical margins is the treatment of choice. A long term follow-up is also advised to the patients.

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Legends to Figures

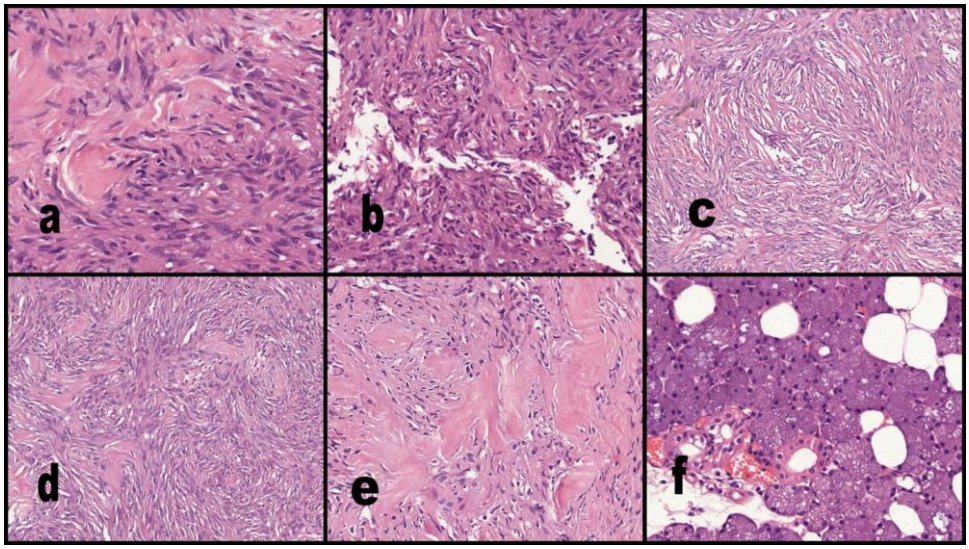

Figure 1 (a) Photomicrograph shows alternate hypercellular and hypocellular areas of tumour tissue (HE×400). **(b)** Photomicrograph shows staghorn-like appearance of a blood vessel (HE×400). **(c)** Tumour cells forming a fascicle (HE×200). **(d)** Photomicrograph shows storiform pattern of tumour cells (HE×200). **(e)** Photomicrograph shows fibrous tumour stroma with areas of collagen deposition (HE×200). **(f)** Photomicrograph shows normal serous salivary gland acini and ducts (HE×400).

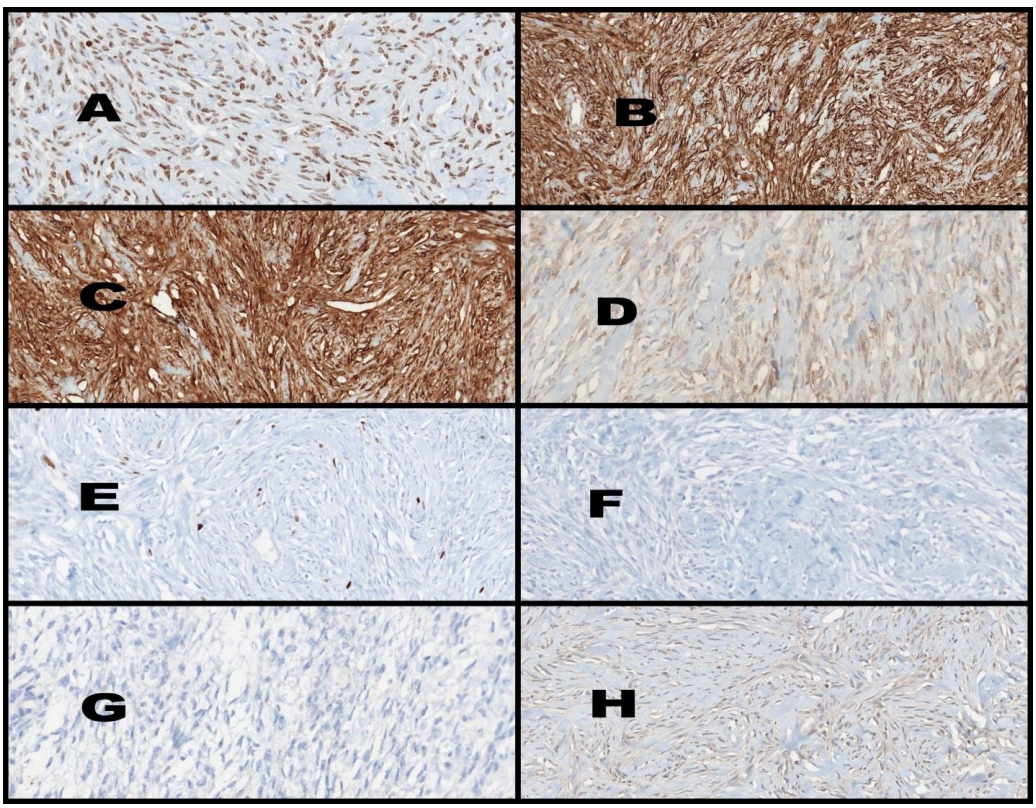

Figure 2 (A) Anti-STAT6 antibody stained nuclei of tumour cells as positive. **(B)** Tumour cells showed strong diffuse membranous positivity with anti-CD34 antibody. **(C)** Anti-CD99 antibody showed diffuse strong membranous positivity of the tumour cells. **(D)** Anti-Bcl2 antibody stained nuclei as well as the cytoplasm of tumour cells. **(E)** Tumour showed low proliferative activity with anti-Ki67 antibody (<4% of nucleated cells). **(F)** Anti-PCK antibody failed to stain the tumour cells. **(G)** Tumour cells showed negative reaction with anti-GFAP antibody. **(H)** Anti-myogenin antibody showed aberrant positivity of the tumour cells.