

Solitary fibrous tumor of parotid gland: a case report and short review of literature

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Abstract. – OBJECTIVE: This case report represents a solitary fibrous tumor, which is a very rare neoplasm in the parotid gland.

CASE PRESENTATION: 80-year-old Caucasian female patient with palpable, immobile, painless, slow growing, semi-rigid-elastic neoplasm in the left parotid gland, that was existing for four months. There were no signs of inflammation and facial paralysis. The tumor was initially interpreted as a salivary gland neoplasm of uncertain origin. Fine needle aspiration was performed 2 times; however, the precise diagnosis was challenging. The tumor was excised, regional lymphadenectomy was performed. Morphological and immunophenotypical findings were consistent with solitary fibrous tumor of parotid gland. Currently, the patient is under regular follow-up period at 9 months with no evidence of metastasis or recurrence.

CONCLUSIONS: Although very rare, solitary fibrous tumor (SFT) should be suspected in cases of slow growing, solid, well-defined nodules of the parotid gland. The SFT of the parotid gland are usually benign tumors, however there is a low risk of recurrency and malignant behavior with metastatic potential. Complete resection of the tumor should be performed since it is crucial for management without any recurrence.

Key Words:

Solitary fibrous tumor, Parotid gland, Salivary gland, Fine needle aspiration cytology, Differential diagnosis.

Introduction

Solitary fibrous tumor (SFT) is a mesenchymal spindle cell neoplasm with fibroblastic differentiation that can occur anywhere in the body¹⁻³.

SFT is mostly seen in the pleura and peritoneum². Most of these tumors have benign features, but some have malignant potential². SFT is not common in the head and neck region, and involvement of the parotid gland is a rarer phenomenon with limited number of cases reported in the literature³⁻⁷. The clinical characteristics of SFT is a painless, slow-growing, palpable, with clearly defined boundaries with no signs of invasion to the neighboring structures⁷.

The aim of this study is to report diagnostics and management of a patient with SFT of parotid gland. The patient represented had a 3.5 cm palpable, immobile, semi-rigid tumor adhering to the deep planes in the parotid gland. The lesion was surgically excised by asportation of neoformation on left parotid gland and regional parotid lymph nodes dissection. The patient was followed-up for 9 months, with no signs of complications and recurrence. The clinical, histopathological, immunohistochemical diagnosis and treatment aspects of the neoplasm are discussed in this paper.

Case Presentation

This case report represents the management of an 80-year-old female Caucasian patient with uni-lateral swelling over the parotid area that was referred to the Maxillo-facial Department at Ospedale Maggiore Policlinico of Milan, Italy. The study protocol followed the principles laid down in the Declaration of Helsinki on medical protocol. A signed informed consent agreement form was obtained from the patient.

In brief, the patient was otherwise healthy but had complaints of clearly visible, asymptomatic, non-tender slowly enlarging swelling over the parotid area of 4 months duration on the left side of the face. Patient did not have any pain, discomfort, or any watery discharge from the lesion. Physical examination showed an approximately 3.5 cm palpable, immobile, semi-tense/elastic mass adhering to the deep planes in the parotid region. The patient represented no signs of OSAS or facial paralysis. The overlying skin was normal and did not show any signs of inflammation with no palpable laterocervical lymph nodes in the neck area. However, there was a very slight enlargement at the superficial parotid lymph nodes.

For diagnosis, the patient was referred to Radiology department for sonogram, and fine needle aspiration cytology (FNAC) was scheduled.

Ultrasonography

Ultrasonography of the neck area revealed a 4×3 cm mixed component lesion over the left parotid area.

First Fine Needle Aspiration (FNA)

A total of four specimens were collected and were fixed in a tube containing 35 ml of 95% ethyl alcohol. Two cytological specimens were prepared with MGG stain and two with PAP stain.

Microscopic Description of Findings

The smears showed an amorphous background material with few elements showing similar features to epithelioid morphology, associated with some foamy histiocytes.

Cytopathological Diagnosis

The material was inadequate for a definitive diagnostic judgment and a second FNA was planned.

Second FNA

A total of seven specimens were collected and were fixed in a tube containing 35 ml of 95% ethyl alcohol. One cytological specimen was prepared with MGG stain and the other six stained with PAP. The smears were collected in a tube containing 40 ml 95% ethyl alcohol.

Microscopic Description of Findings

Amorphous background material incorporating rare foamy histiocytes, lymphocytes and cell debris.

Cytopathological Diagnosis

Absence of evaluable epithelial component. After an evaluation of the results in the clinic, MRI with contrast and an additional FNA was required.

MRI

Homogeneous and isointense lesion to muscle. On T1- and T2- weighted images; the lesion is heterogeneous and hyperintense. On T2-weighted images strongly enhanced post-gadolinium in areas of mild hyperintensity (Figures 1 and 2).

Surgical Intervention and Histopathologic Findings

Elective enucleoresection of Parotid gland with facial nerve preservation was proposed to the patient. The surgery was planned under general anesthesia. A written informed consent form for the treatment in compliance with the principles of Declaration of Helsinki was provided from the patient before the surgery.

In brief, the surgery included:

- A. Asportation of neoformation on left parotid gland
- B. Asportation of one superficial parotid lymph node.

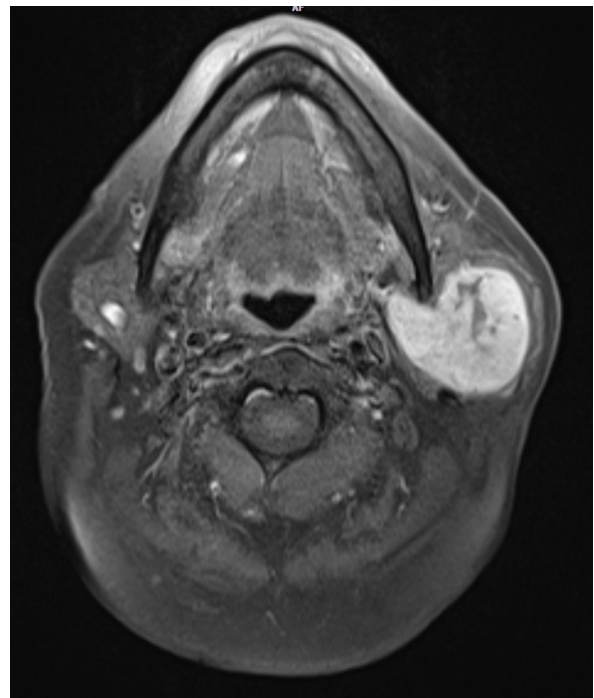


Figure 1. Axial MRI scan view of the patient showing the tumor.

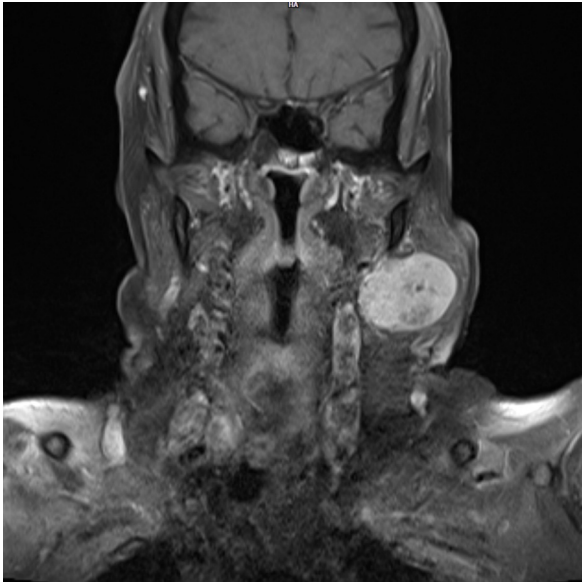


Figure 2. Coronal MRI scan of the patient view showing the tumor.

Surgical Technique

The surgery started with left preauricular and cervical incision, followed by elevation of a superficial cervico-fascial flap elevation between superficial musculo aponeurotic system (SMAS) layer and the parotid fascia until the anterior border. The surgery continued with identification of sternocleidomastoid and digastric muscles, and the identification and preservation of trunk and main branches of the left facial nerve. Enucleoresection of neof ormation of the inferior pole of the left parotid was performed. The resected specimen was sent for further histological examination.

The surgery continued with a careful control of hemostasis, and irrigation with saline. The tissues were sutured and a suction for drainage was placed over SMAS.

Macroscopic Findings

1. Salivary gland with major axis of 3.5 cm showing on the cut surface a 4.5x4x2 cm firm white-yellowish nodule with well-defined margins.
2. Lymph node with major axis of 0.6 cm.

Microscopic Findings

A. The parotid parenchyma showed a proliferation of medium-sized spindle cells with low-grade nuclear atypia, predominantly arranged in storiform pattern with numerous branched

hyalinized vessels and scarce stromal collagen. No necrosis was observed. The mitotic index was focally 5-6 mitosis/10 high-power fields (HPF). The neoplastic cells demonstrated an immunohistochemical expression of STAT6, CD34, CD99 and bcl2, while were negative for Smooth Muscle Actin, Desmin, Cytokeratin AE1/AE3 and ALK1 (Figures 3 and 4).

Morphological and immunophenotypic findings were consistent with solitary fibrous tumor (according to WHO definition), with complete excision.

B. Intraparotid eal lymph node.

Surgical Diagnosis

Benign neof ormation on the left parotid: Solitary fibrous tumor of parotid gland.

During a follow up of 7 months, no recurrence and no adverse event and no intra-/post-operative complications were seen.

Discussion

Solitary fibrous tumor was first described in 1767, followed by the description of Wagner in 1870^{3,8,9}. In 1931, Klemperer and Rabin¹⁰ divided pleural tumors into two classes as diffuse mesotheliomas and localized mesotheliomas or SFT³. Initially, SFT was thought to have a mesothelial origin³. Currently, SFT is considered as a neoplasm of mesenchymal origin, that can be seen in any anatomical site^{3,10-13}. SFT is not very common in the maxillofacial region. Furthermore, SFT

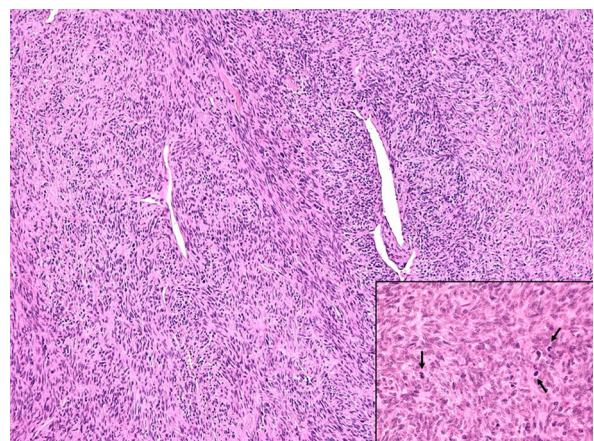


Figure 3. Spindle cell tumor with storiform pattern of growth and "staghorn-like" vessels; mitotic figures are highlighted in the inset (arrows). Staining: hematoxylin-eosin (H&E) magnification 110x, inset H&E, magnification 200x).

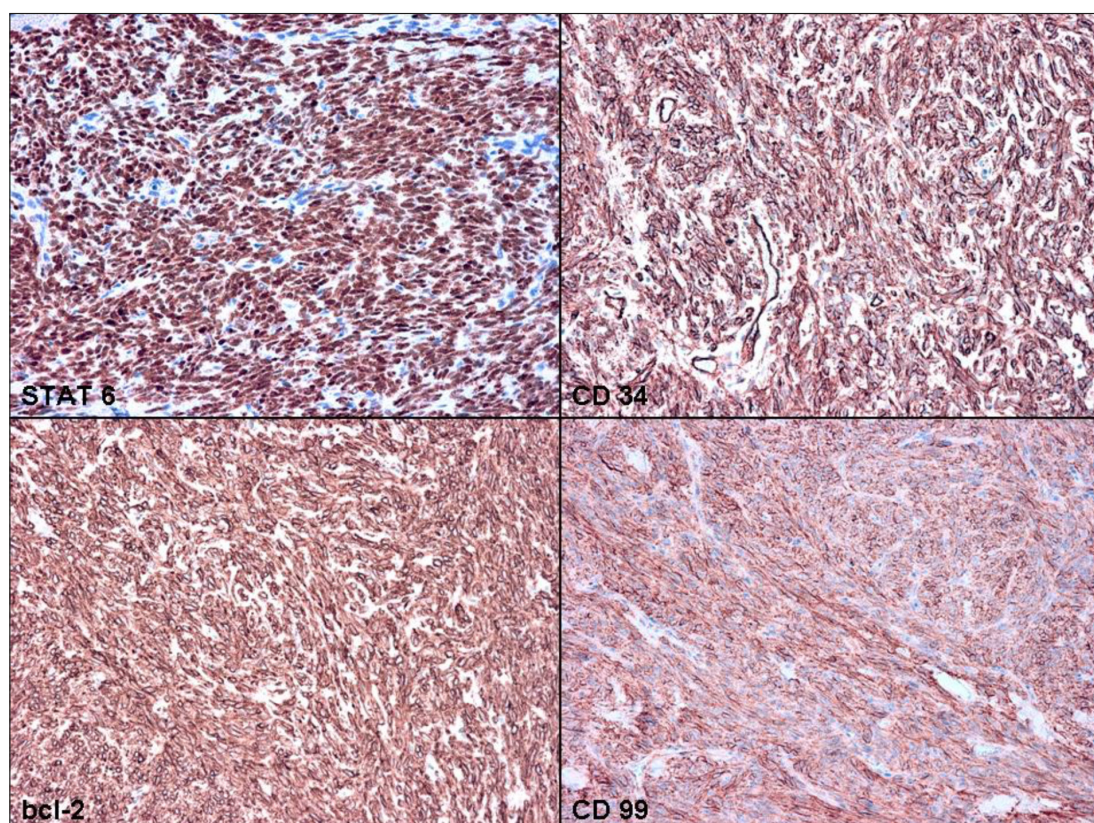


Figure 4. Typical immunophenotype of solitary fibrous tumor with STAT6 (upper left), CD34 (upper right), bcl-2 (lower left) and CD99 (lower right) expression. Magnification 100×.

occurs very rarely in the parotid gland which makes pre-operative diagnosis quite challenging^{14,15}. Although quite infrequent, slow-growing, painless, well-circumscribed nodules in parotid gland should be suspected for SFT in the clinics. In the literature there is a limited information about cytology of parotid SFTs, although majority of the articles are suggesting a spindle cell neoplasm^{3,7}.

Currently, SFT of the parotid gland is considered as a very rare spindle cell neoplasm. In the clinics, these patients can exhibit symptoms of obstructive sleep apnea, which is thought to be related to parapharyngeal extension of the tumor. Additionally, some rare cases reported facial nerve palsy¹⁴. In literature, it was reported that the age ranged from 11 to 79 years³. The median size of tumors range between 1 to 12 cm³. The tumors can be fully or partially encapsulated³. The majority of tumors were described as firm, white-tan or grey masses³. The diagnosis of SFT preoperatively by FNA represents a challenging situation also due to its rarity which rises almost no suspicion initially.

Upon CT examination, SFTs appear as solitary, well-defined masses that are hypointense to muscle and demonstrate heterogeneous contrast enhancement. On magnetic resonance imaging, SFTs are homogeneous and isointense to muscle. On T1- and T2-weighted images; they are heterogeneous and mildly hyperintense. They are also strongly enhanced post-gadolinium in areas of mild hyperintensity on T2-weighted images^{14,16}. In this case, the initial diagnosis was not possible with FNA and after a re-evaluation visit of the FNA findings in the clinic, MRI with contrast and an additional FNA was required. As a result, the MRI findings of this case was matching with the diagnostic features that are reported in the literature.

The typical histologic features of SFT include a variable proliferation of bland ovoid to spindle cells, arranged in fascicular to patternless architecture, along with scar or keloid-like dense collagen deposition and a rich vascular plexus with so called “staghorn” pattern.

The most sensitive and specific immunohistochemical marker of SFT is STAT6, but also CD34, CD99 and bcl2 are expressed in most cases.

Solitary fibrous tumors of the parotid gland usually exhibit benign features; however, they should be regarded as potentially malignant, because there are no standardized criteria for classifying SFT^{14,17}. To evaluate its behavior, the morphological and histological assessments are crucial. The frequency of malignancy for SFTs that is reported in the literature, shows a range between 10 % and 37 %, in any case complete resection should be considered to eliminate any potential risks¹⁷⁻¹⁹. Currently, for the management of SFT, a resection surgery with wide healthy margins is considered as the gold standard¹⁷. Morphologically, malignancy must be suspected in presence of cytologic atypia, necrosis and increased mitotic index (>4 mitosis/10HPF).

In our case, the immunofenotypical profile showed STAT6 +, CD34 +, CD99 +, bcl2 +, Smooth Muscle Actin -, Desmin -, Citokeratin AE1/AE3 -, ALK -. Morphological and immunophenotypic findings were consistent with solitary fibrous tumor (according to WHO definition) with clean resection margins^{14,17}.

The findings in this case report of parotid SFT, the size of the tumor, duration of symptoms, and immunohistology findings were consistent with previous reports. At present, in the English literature only thirty-seven cases of SFT in the parotid gland have been reported, including this case report^{5-8,14,18,20-34}. Although, SFT is infrequently found in parotid region, it should be always suspected in cases of painless, slow-growing, palpable tumors, with clearly defined boundaries. Limitations of this work include a relatively short follow up period of the patient.

Conclusions

Although very rare, SFT should be suspected in cases of slow growing, solid, well-defined nodules of the parotid gland. Fine needle aspirations can be not sufficient to reach to a final diagnosis in these cases. Additional analyses including MRI and immunochemistry should support the results to reach a conclusion.

The SFT of the parotid gland are usually benign tumors, however there is a low risk of recurrence and malignant behavior with metastatic potential. Complete resection of the tumor should be performed since it is crucial for management without any recurrence.

Conflict of Interest

The authors declare no potential conflicts of interest with respect to the authorship and/or publication of this article.

Acknowledgements

None.

Ethics Approval

Not applicable for a case report.

Funding Statement

This study was funded by Italian Ministry of Health. Current research IRCCS (Fondazione IRCCS Ca' Granda Ospedale Maggiore Policlinico).

Authors' Contribution

F.G., C. Ma., M.M., G.B., M.D.F., A.B., F.R.P.B., A.R., A.B.G. and D.S.R. conceived and designed the analysis. Databases were searched and data was collected by F.G., C.M. F.R.P.B., A.R., C. Mo. and D.S.R. All the authors contributed on analysis and interpretation of data for the work. F.G. drafted the work and wrote the manuscript with input from all authors. F.G., C. Ma., M.D.F., A.B., A.B.G., M.M., A.R., G.B., F.R.P.B., C. Mo., and D.S.R. revised the work critically for intellectual content. Integrity of the work was appropriately investigated and resolved by all authors. All authors contributed and approved equally to the final version of the manuscript.

Informed Consent

A signed informed consent agreement form was obtained from the patient.

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