

J. Sato · K. Asakura · Y. Yokoyama · M. Satoh

Solitary fibrous tumor of the parotid gland extending to the parapharyngeal space

Received: 28 May 1997 / Accepted: 25 August 1997

Abstract Solitary fibrous tumors (SFT) arise in the pleura and less commonly in extrapleural sites. Head and neck regions have included the nose and paranasal sinuses, soft palate, epiglottis, thyroid, parotid and submandibular glands, as well as the infratemporal fossa and parapharyngeal space. We report a case of SFT arising from the parotid gland and extending to the parapharyngeal space. To our knowledge, this is the fourth case of SFT originating from the parotid gland and is the largest of its kind among the extrapleural lesions described. The characteristics revealed by computed tomography and magnetic resonance imaging are presented.

Key words Parotid gland · Solitary fibrous tumor · Magnetic resonance imaging

Introduction

The solitary fibrous tumor (SFT) was first described in 1931 [10]. These tumors often arise in the pleura [1], but can be found in various head and neck sites. Although rare, these locations have included the infratemporal fossa [12], parapharyngeal space [13], nose and paranasal sinuses [3, 16], soft palate [14], epiglottis [13], as well as the thyroid [15], parotid [5, 8, 14] and submandibular gland [7, 14]. We now report a case of SFT arising from the parotid gland and extending to the parapharyngeal space.

Case report

The patient was a 52-year-old Japanese office worker who first noticed a small mass in his left pre-auricular region in 1992. This lump grew slowly without causing distress, but symptoms of obstructive sleep apnea, dysphagia and altered speech were noted by his family from the beginning of 1996, after which he was seen at Sapporo Medical University Hospital on February 28, 1996. Physical examination showed a painless, firm 65-mm mass in the left parotid region. The facial nerve and the other cranial nerves were intact. A diffuse submucosal swelling involving the left antero-lateral wall of the nasopharynx and oropharynx was seen and a 20 mm × 8 mm area of mucosal ulceration was found at the posterior end of the left upper givivo-buccal sulcus.

CT showed a large dumbbell-shaped and well-encapsulated mass in the left parotid and parapharyngeal region (Fig. 1). The surface of the tumor was slightly enhanced with contrast medium, and the central portion was irregularly enhanced. There were no radiolucent areas between the tumor and parotid. The mass extended from the skull base and had created a bone defect to the oropharynx. The left pterygoid process, posterior wall of the maxillary antrum and mandibular rams were eroded. The left great vessels were dislocated posteriorly, but without signs of tumor invasion.

On MRI (Fig. 2), the mass showed a low-intermediate signal intensity on T1-weighted images, low signal intensity on T2-weighted images and heterogeneous enhancement on Gadolinium-enhanced magnetic resonance imaging (GadMRI). Further imaging with ^{99m}Tc-pertechnetate showed a slight uptake into the tumor, although this uptake was less than that seen in a Warthin's tumor. No uptake of gallium citrate was seen.

On April 15, 1996, tumor resection had been performed by an external approach. Left superficial parotidectomy was performed first. The deep lobe of the parotid gland was then completely replaced by the tumor, but facial nerve invasion had not occurred. A midline mandibular osteotomy was performed and a large mass extending to the parapharyngeal space was resected by using a mandibular swing method (Fig. 3). As the tumor surface was smooth and clearly defined, it could be bluntly dissected from surrounding tissue. After removing the tumor, a 25-mm round-shaped bone defect was left at the skull base. However the dura, great vessels, cranial nerves, pharyngeal constrictor muscles and overlying mucosa had not been invaded by the tumor and remained intact after the dissection. The left mandibular ramus at the root of its articular process had been thinned by compression of the tumor but was otherwise intact. A fat pad was obtained from the abdominal wall and was placed in the skull base defect to fill the postoperative dead space. The postoperative course was good, and there

J. Sato (✉) · K. Asakura · Y. Yokoyama
Department of Otolaryngology, School of Medicine,
Sapporo Medical University, S1W16, Chuo-Ku,
Sapporo 060, Japan

M. Satoh
Department of Pathology, Sapporo Medical University,
Sapporo, Japan

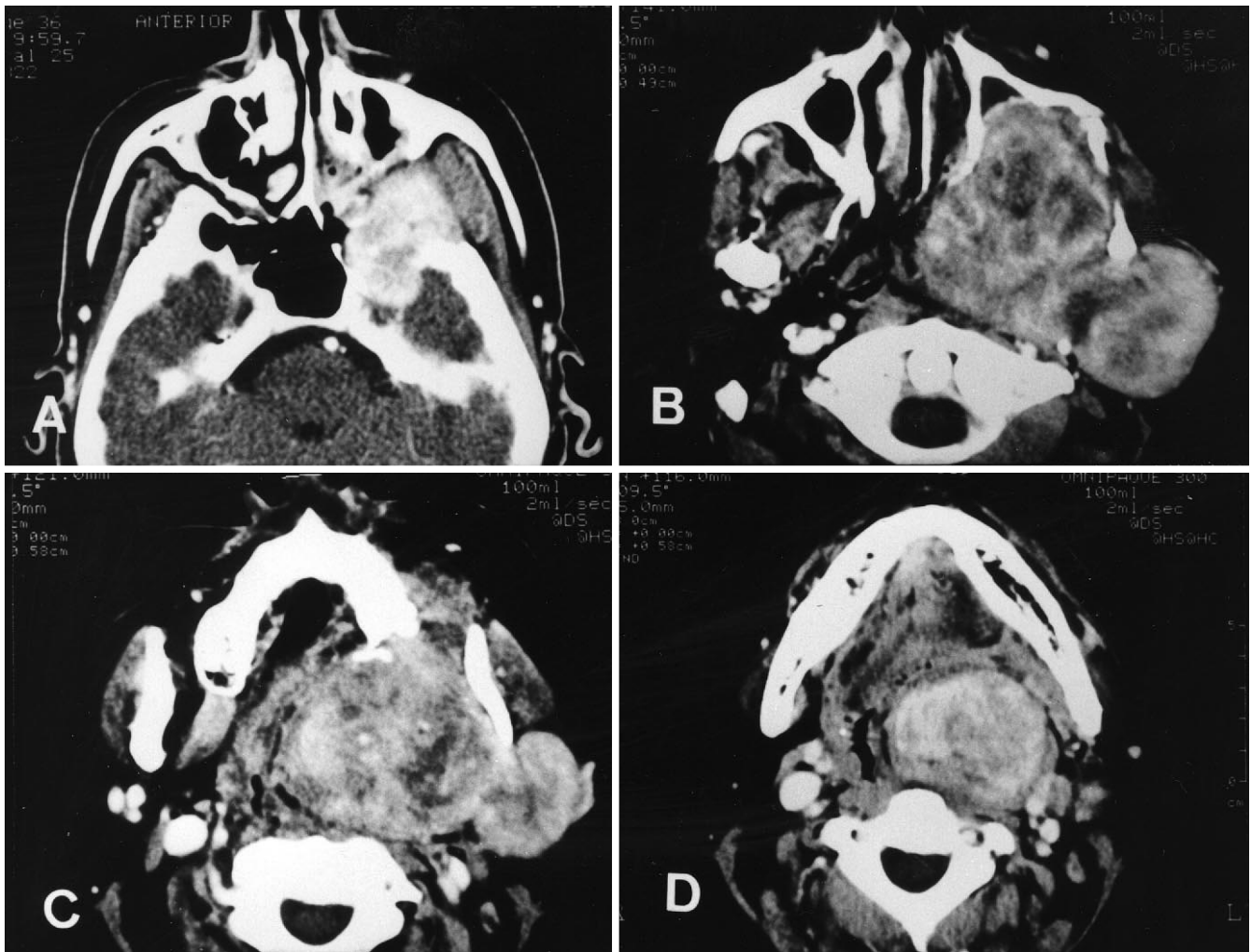


Fig. 1 Axial computed tomography showing tumor at the levels of skull base (A), external auditory canal (B), maxilla (C) and mandible (D)

has been no evidence of tumor recurrence 1 year after the operation.

Grossly, the tumor was well encapsulated and measured 12 cm × 10 cm × 5.5 cm, weighting 310 g (Fig. 4). The cut surface was firm and pale in color. Microscopic examination showed a growth of spindle-shaped cells in a heavy collagenized stroma without epithelial components (Fig. 5 A). Non-structured bundles of cells in storiform patterns were observed. No atypia, mitosis or necrosis was found. An immunohistochemical study revealed that tumor cells were positive for vimentin and CD34, but negative for desmin, S-100 and smooth muscle actin (Fig. 5 B).

Discussion

To our knowledge, our case of SFT is the fourth case to originate from the parotid gland [8, 14] and is the largest of its kind among the extrapleural lesions described. Although rare cases of SFT may present with systemic symptoms, such as hypoglycemia, arthralgias, osteoarthropathy

and finger clubbing, most cases are discovered through an incidental radiological finding or as the result of symptoms related to a mass effect [11].

In our case, CT and MRI showed a clearly marginated dumbbell-shaped mass extending from the deep lobe of the left parotid gland to the parapharyngeal space with skull-base bone destruction. That no fatty tissue image was seen between the tumor and parotid tissue suggested that the tumor had originated from the gland per se and had then extended to the parapharyngeal space. On CT examination, the superficial portion of the tumor was slightly enhanced with contrast medium, while the central portion was irregularly enhanced. No high-density area was presented to suggest calcification.

On MRI, the appearance of our patient's tumor conformed to the characteristics of fibrous tissue, exhibiting a low-intermediate signal on both T1- and T2-weighted images. Although it has been reported that high-grade malignant neoplasms of the parotid gland sometimes show low intensity on T1- and T2-weighted images [2], most parotid gland and parapharyngeal tumors show a low signal intensity on T1-weighted images and a high signal intensity on T2-weighted images. The low mobile proton density of the SFT is thought to be responsible for the sig-

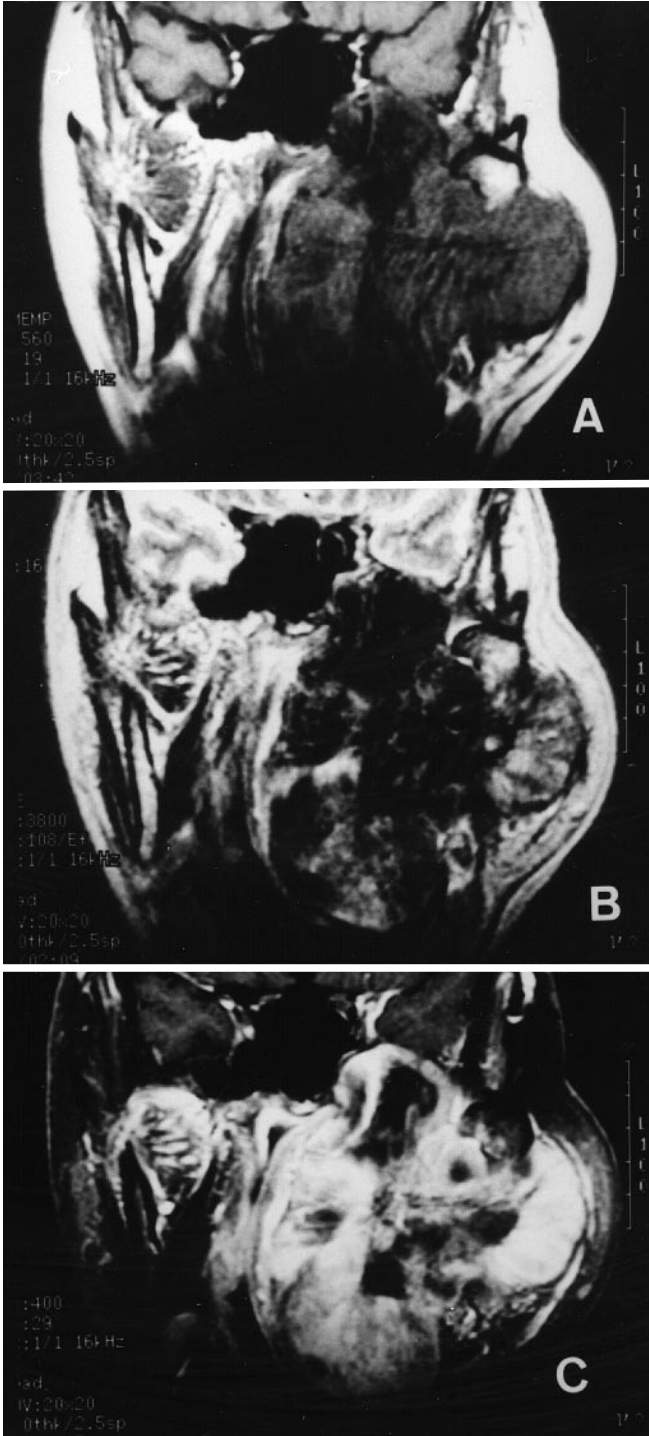


Fig.2A–C Coronal magnetic resonance imaging (MRI). **A** T1-weighted scans show a homogeneous low intensity tumor. **B** T2-weighted scans show the tumor with a heterogeneous low intensity. **C** Gadolinium-enhanced MRI. The tumor shows high heterogeneous enhancement

nal intensity seen on the T2-weighted image. The GadMRI showed a mixed but predominantly high-contrast image in our case. These findings may be characteristic of the solitary fibrous tumor [9, 11].

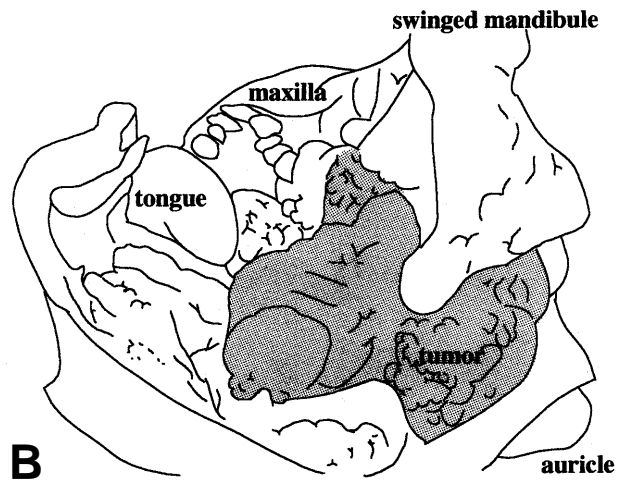
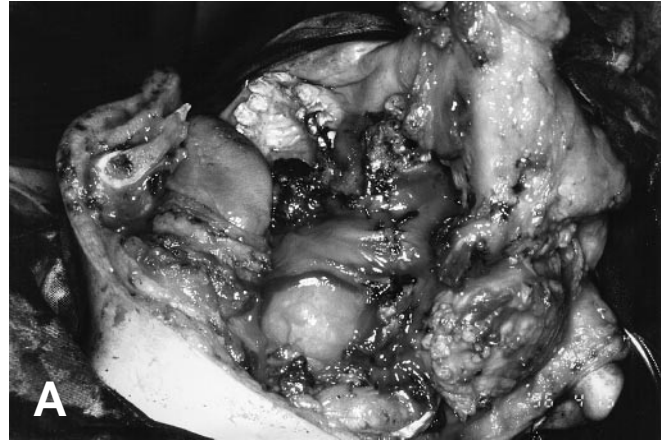


Fig.3 A Intraoperative situation after midline mandibular osteotomy and rotation laterally and superiorly (mandibular swing method). **B** Schematic demonstration of operation findings

The pathological findings in our case revealed non-structured bundles of cells in a storiform configuration and a rich fibrous matrix on light microscopy [5, 8, 14, 15]. This appearance is the so-called patternless pattern. Histologically, SFT can be difficult to distinguish from other spindle-cell tumors, although SFT has been reported to be reaction-positive for CD34 and vimentin [8]. In our case, immunohistochemical study showed positive staining of spindle cells for both vimentin and CD34, but negative for desmin, S-100 and smooth muscle actin.

Most SFTs are clinically benign tumors, and the recommended management is complete surgical resection. In our case, the tumor was resected successfully by an external approach, and the patient has now been free of recurrence for 1 year since the operation. Previous reports have indicated that 13–37% of SFTs were associated with local recurrence or histological malignancy [4, 6]. Goodlad and Fletcher [6] described a case with tumor recurrence 31 years after surgical excision. Therefore, long-term follow-up is very important even in cases in which pathological findings suggest that the SFT is benign.

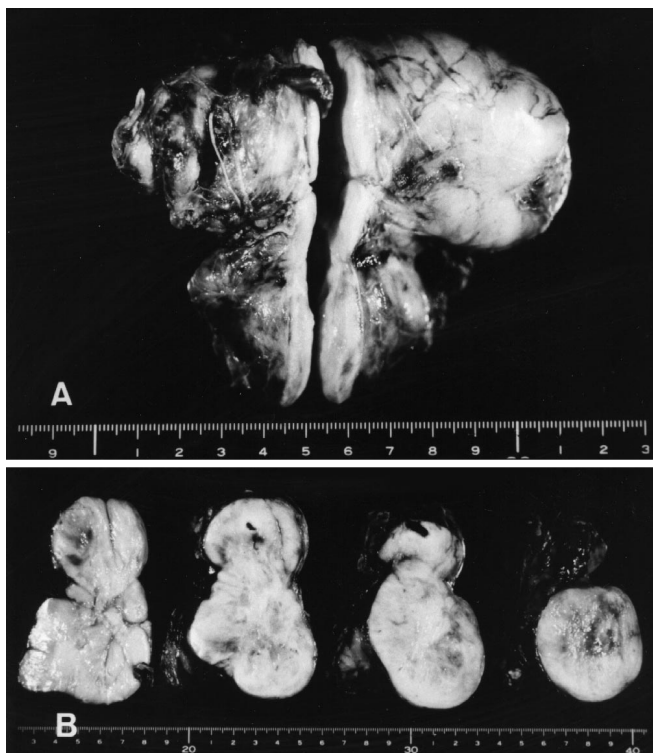


Fig. 4 **A** Gross appearance of excised tumor. **B** Cut surface of tumor

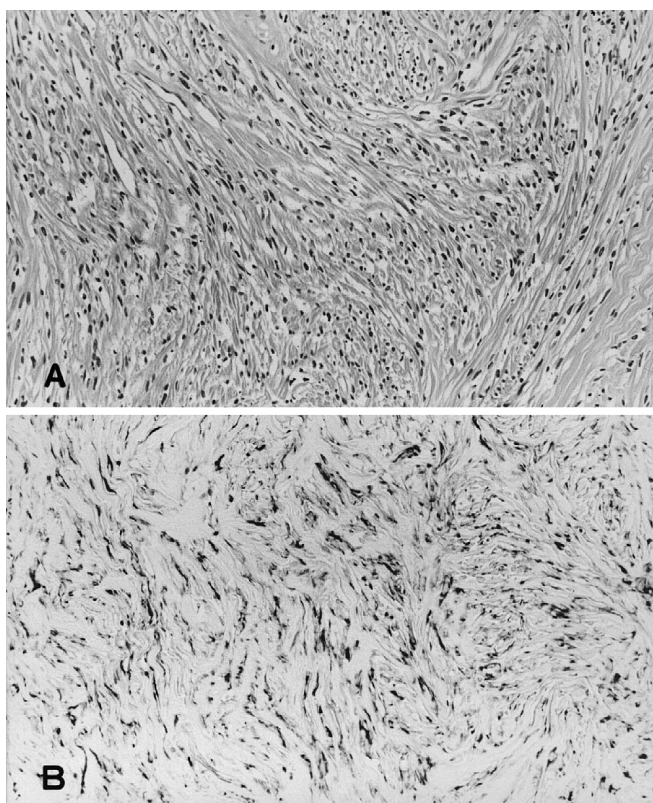


Fig. 5 **A** Hematoxylin and eosin-stained section of tumor showing "pattern-less" pattern. **B** Large numbers of tumor cells are positive for CD34. $\times 240$

Acknowledgements The following members of the Department of Otolaryngology, Sapporo Medical University participated in the present report: Drs. Hideki Ogasawara, Toshinori Matsui, Takayuki Kozawa, and Akikatsu Kataura.

References

1. Briselli M, Mark EJ, Dickersin GR (1981) Solitary fibrous tumors of the pleura: eight new cases and review of 360 cases in the literature. *Cancer* 47: 2678–2689
2. Byrne MN, Spector JG, Gravin CF, Gado MH (1989) Preoperative assessment of parotid masses: a comparative evaluation of radiologic techniques to histopathologic diagnosis. *Laryngoscope* 99: 284–292
3. el-Naggar AK, Weard RM, Ro JY, Ayala AG, Ordonez NG (1987) Fibrous tumor with hemangiopericytic pattern, so-called "localized fibrous tumor of the pleura." *Lab Invest* 56: 21
4. England DM, Hochholzer L, McCarthy MJ (1989) Localized benign and malignant fibrous tumors of the pleura. A clinicopathologic review of 223 cases. *Am J Surg Pathol* 13: 640–658
5. Ferreiro JA, Nascimento AG (1996) Solitary fibrous tumour of the major salivary glands. *Histopathology* 28: 261–264
6. Goodlad JR, Fletcher CD (1991) Solitary fibrous tumour arising at unusual sites: analysis of a series. *Histopathology* 19: 515–522
7. Gunhan O, Yildiz FR, Celasun B, Onder T, Finci R (1994) Solitary fibrous tumour arising from sublingual gland: report of a case. *J Laryngol Otol* 108: 998–1000
8. Hanau CA, Miettinen M (1995) Solitary fibrous tumor: histological and immunohistochemical spectrum of benign and malignant variants presenting at different sites. *Hum Pathol* 26: 440–449
9. Harris GN, Rozenshtein AM, Schiff MJ (1995) Benign fibrous mesothelioma of the pleura: MR imaging findings. *AJR Am J Roentgenol* 165: 1143–1144
10. Klemperer P, Rabin C (1931) Primary neoplasms of the pleura. *Arch Pathol* 11: 385–412
11. Padovani B, Mouroux J, Raffaelli C, Huys C, Chanalet S, Michiels JF, Brunner P, Bruneton JN (1996) Benign fibrous mesothelioma of the pleura: MR study and pathologic correlation. *Eur Radiol* 6: 425–428
12. Rayappa CS, McArthur PD, Gangopadhyay K, Antonius JI (1996) Solitary fibrous tumour of the infratemporal fossa. *J Laryngol Otol* 110: 594–597
13. Safneck JR, Alguacil Garcia A, Dort JC, Phillips-SMT (1993) Solitary fibrous tumour: report of two new locations in the upper respiratory tract. *J Laryngol Otol* 107: 252–256
14. Suster S, Nascimento AG, Miettinen M, Sickel JZ, Moran CA (1995) Solitary fibrous tumors of soft tissue. A clinicopathologic and immunohistochemical study of 12 cases. *Am J Surg Pathol* 19: 1257–1266
15. Taccagni G, Sambade C, Nesland J, Terreni MR, Sobrinho Simoes M (1993) Solitary fibrous tumour of the thyroid: clinicopathological, immunohistochemical and ultrastructural study of three cases. *Virchows Arch* 422: 491–497
16. Zukerberg LR, Rosenberg AE, Randolph G, Pilch BZ, Goodman ML (1991) Solitary fibrous tumor of the nasal cavity and paranasal sinuses. *Am J Surg Pathol* 15: 126–130