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A Case of Solitary Fibrous Tumor of the Parotid Gland: Review of the Literatures

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Kumagai, M., Suzuki, H., Takahashi, E., Matsuura, K., Furukawa, M., Suzuki, H. and Tezuka, F. *A Case of Solitary Fibrous Tumor of the Parotid Gland: Review of the Literatures.* Tohoku J. Exp. Med., 2002, **198** (1), 41-46 —— We report a rare case of solitary fibrous tumor of the parotid gland. A 47-year-old woman presented with a 3-year-history of left-sided subauricular swelling. Computed tomographic scans and magnetic resonance images revealed a well-defined and dumbbell-shaped mass, measuring about 30 mm in its greatest dimension, in the left parotid gland. Because the tumor occupied both superficial and deep lobes of the gland, she underwent total parotidectomy with preservation of the facial nerve. The microscopic finding showed short-spindle and ovoid cells arranged in a haphazard pattern with interspersed thin collagen fibrils. Immunohistochemically, the tumor cells were strongly positive for CD34, bcl-2 and vimentin, whereas stains for S-100, cytokeratin, smooth muscle actin, collagen type IV and CD117 (KIT) were negative. On the basis of these findings, the tumor was diagnosed as solitary fibrous tumor. Her post-operative course was uneventful, and she is currently free from disease 14 months after surgery. Diagnosis, clinical behavior and treatment of solitary fibrous tumor are reviewed from perusal of the literature. ———— solitary fibrous tumor; parotid gland; immunohistochemistry; CD34

Solitary fibrous tumor (SFT) is a soft tissue neoplasm generally associated with serosal surfaces, most commonly, the visceral pleura. Since Klemperer and Rabin (1931) first described SFT of the pleura, SFTs have also been reported in extrapleural sites unrelated to serosal surfaces. In the head and neck, SFTs have been documented in the nose and paranasal sinuses (Zukerberg et al. 1991), nasopharynx (Mentzel et al. 1997), larynx (Safneck et al. 1993; Benlyazid et al. 1998), oral cavity (Alawi et al. 2001), orbit (Havlik et al. 2000), paraphar-

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yngeal space (Gangopadhyay et al. 1996), thyroid (Kie et al. 1997) and salivary glands (Ferreiro and Nascimento 1996). To the best of our knowledge, 9 cases of SFTs arising from the major salivary glands have so far been reported in the English literatures: 5 from the parotid gland (Hanau and Miettinen 1995; Suster et al. 1995; Ferreiro and Nascimento 1996; Sato et al. 1998; Mohammed et al. 2001), 2 from the submandibular gland (Ferreiro and Nascimento 1996; Guarino et al. 1998) and 2 from the sublingual gland (Ferreiro and Nascimento 1996; Gunhan et al. 1994). Here, we report the sixth case of SFT of the parotid gland.

Case Report

A 47-year-old woman who had complained of left-sided subauricular swelling for three years was referred to our clinic on June 28, 2001. She had a past history of uterine myoma, and no particular family history of illness. On physical examination, there was an elastic soft, round and mobile mass in her left subauricular region. Her facial movement was intact. No abnormal findings were noted in the oral cavity, nose, pharynx or larynx. Computed tomographic scans and magnetic resonance images (Fig. 1) revealed a well-defined and slightly-enhanced tumor, measuring about 30 mm in its greatest dimension, in the left parotid gland. The tumor was dumbbell-shaped and extended to the parapharyngeal space. It showed isointensity and slightly high intensity in T1- and T2-weighted magnetic resonance images, respectively. She was clinically diagnosed as a left-sided parotid tumor. Fine needle aspiration biopsy showed class III (borderline malignancy). Because the tumor occupied both superficial and deep lobes of the parotid gland, she underwent total parotidectomy with preservation of the facial nerve on September 21, 2001. She did not manifest facial nerve palsy, and had an uneventful clinical course post-operatively.

The resected tumor was $42 \times 35 \times 32$ mm in size and was well-circumscribed. Its cross-section showed a firm, whitish and homogenous appearance. The microscopic finding was characterized by the proliferation of short-spindle and ovoid cells arranged in a haphazard pattern with interspersed thin collagen fibrils (Fig. 2). The mitotic rate was 5/10 high power fields, however, no necrotic lesions, extracapsular invasion or cell atypia were observed. The tumor

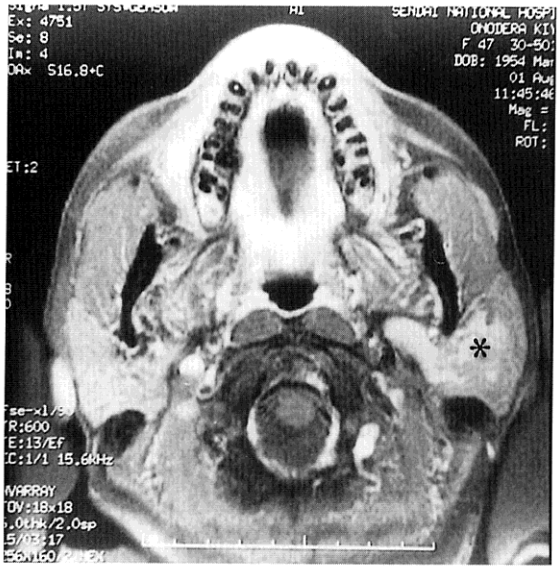

Fig. 1. Gadolinium-enhanced T1-weighted magnetic resonance image (axial section).
Note that the tumor (*) of the left parotid gland is extended to the parapharyngeal space.

was, therefore, considered to be essentially benign. Immunohistochemically, the tumor cells were strongly positive for CD34 (Fig. 3), bcl-2 and vimentin. Stains for S-100, cytokeratin, smooth muscle actin, collagen type IV and CD117 (KIT) were negative. On the basis of these findings, the tumor was diagnosed as SFT. No additional treatment was given to her because the operative and histopathological findings indicated that the tumor was completely resected and was essentially benign. The patient is currently free from disease 14 months after surgery.

Discussion

The histogenesis of SFT has been controversial. This neoplasm was formerly referred to as fibrous mesothelioma, solitary fibrous mesothelioma, localized fibrous mesothelioma, localized mesothelioma, and so forth (Chan 1997). However, SFT is immunophenotypically distinct from mesothelioma, and is thought to be derived from fibroblastic or myofibroblastic cells (Dervan et al. 1986; Morgan and Smoller 2000). In the salivary glands as well, SFT presumably originates from

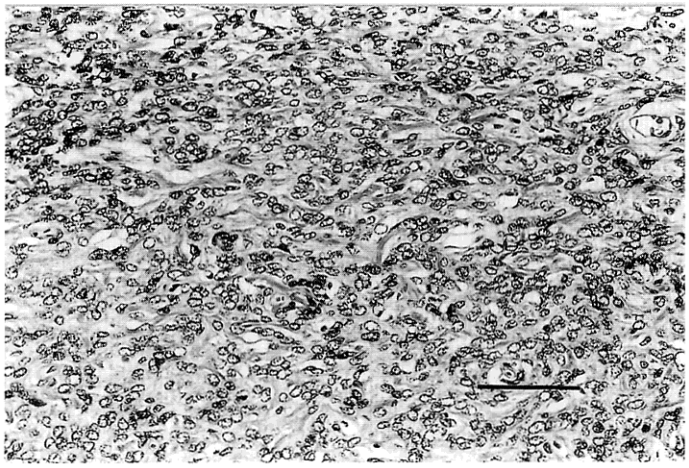

Fig. 2. Microscopic finding of the tumor showing short-spindle and ovoid cells arranged in a haphazard pattern with interspersed thin collagen fibrils. Scale bar = 100 μm. (hematoxylin & eosin stain)

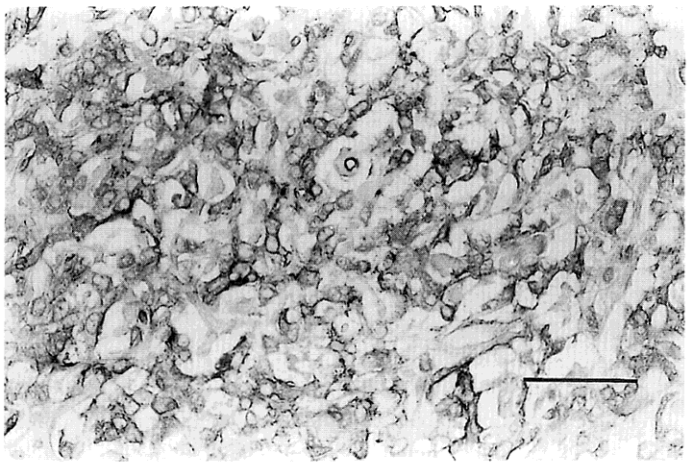

Fig. 3. CD34 immunohistochemical staining showing the strong immunoreactivity of the tumor cells. Scale bar = 50 μm.

such mesenchymal cells of the parenchyma. The histological appearance of SFT consists of short-spindle or ovoid cells in various patterns such as haphazard, storiform or fascicular arrangement with interspersed thin or thick collagen fibrils (Chan 1997). The diagnosis of SFT is often difficult because there are a number of other soft tissue neoplasms that show similar histological findings. Immunohistochemical examinations are, accordingly, required to differentiate SFT from other analogous tumors. The typical immunohistochemical profile of SFT represents consistent immunoreactivity for CD34, bcl-2 and vimentin, and negative staining for S-100, cytokeratin, desmin, smooth muscle actin and CD117 (Chan 1997; Sarlomo-Rikala et al. 1998; Alawi et al. 2001). Among these molecules, CD34, a 100 kDa transmembrane glycoprotein expressed in hematopoietic progenitor cells, is the most important and sensitive marker for the diagnosis of SFT (Westra et al. 1994).

Not only SFT but also several other soft tissue tumors possess immunoreactivity for CD34. Neurofibroma and schwannoma may be reactive for CD34, however, these tumors are also positive for S-100 while SFT is not (Alawi et al. 2001). Dermatofibrosarcoma protuberans is another CD34-positive soft tissue tumor, but it is often bcl-2 negative (Alawi et al. 2001). Gastrointestinal stromal tumor also represents histological and immunohistochemical similarities to SFT. The expression of CD117, a transmembrane growth factor receptor that is the product of the c-kit gene, is one of the criteria for the diagnosis of gastrointestinal stromal tumor (Rader et al. 2001), whereas SFT lacks CD117 (Sarlomo-Rikala et al. 1998). Hemangiopericytoma is one of the most difficult tumors to distinguish from SFT because of their striking histological and immunohistological similarities. Although CD34 expression has been reported in hemangiopericytoma, the staining is patchy in distribution and weak in intensity, which is in contrast to the strong and diffuse staining in SFT (Ferreiro and Nascimento 1996).

The present case showed a strong immunoreactivity for CD34 and bcl-2, and no reactivity for S-100 and CD117, confirming the diagnosis of SFT.

According to the literature, 5 to 23% of pleural SFTs behave in a clinically malignant manner, as demonstrated by local invasion, local recurrence, intrathoracic spread or distant metastasis (Briselli et al. 1981; England et al. 1989; Chan 1997; Alawi et al. 2001). Atypical histological findings include ambiguous circumscription, nuclear pleomorphism, high cellularity, a high mitotic rate and necrosis. Neverthe-

TABLE 1. *Reported cases of SFTs of the parotid gland*

Age	Sex	Side	Size (mm)	Therapy	Follow-up	Author (year)
73	Female	Left	35×25×25	Local excision	No recurrence at 4 years	Hanau and Miettinen (1995), Suster et al. (1995)
78	Female	Right	10	Superficial lobectomy	No recurrence at 1 year	Suster et al. (1995), Ferreiro and Nascimento (1996)
46	Male	Left	44	Superficial lobectomy	No recurrence at 1 month	Ferreiro and Nascimento (1996)
52	Male	Left	120×100×55	Total parotidectomy with midline mandibulotomy	No recurrence at 1 year	Sato et al. (1998)
42	Female	Right	22	Local excision	No recurrence at 3 months	Mohammed et al. (2001)
47	Female	Left	42×35×32	Total parotidectomy	No recurrence at 14 months	Present case (2002)

less, no unifying histological criteria reliably and consistently predict the malignancy of SFT. On the other hand, most extrathoracic SFTs are clinically benign (Chan 1997; Alawi et al. 2001). The six reported cases of SFTs of the parotid gland including the present one are summarized in Table 1. In none of the cases, recurrence was seen in the follow-up periods of 1 month to 4 years, with an average of 15 months, after surgery.

While many authors described that complete surgical resection is the only and best treatment for SFT, Goodlad and Fletcher (1991) obtained good results with radiotherapy and chemotherapy against residual tumor after incomplete resection of SFTs. Local recurrence may occur long after surgery. Goodlad and Fletcher (1991) documented a case of SFT of the pleura that had recurred 31 years after complete surgical resection. Long-term follow-up is, therefore, mandatory, even if the tumor appears to be clinically benign and completely resected. The present case meets such circumstances, and is to be under close follow-up hereafter.

References

Alawi, F., Stratton, D. & Freedman, P.D. (2001) Solitary fibrous tumor of the oral soft tissues: a clinicopathologic and immunohistochemical study of 16 cases. *Am. J. Surg. Pathol.*, **25**, 900-910.

Benlyazid, A., Lescanne, E., Lefrancq, T., Fetissoff, F. & Beutter, P. (1998) Solitary fibrous tumor of the larynx: report of a case. *J. Laryngol. Otol.*, **112**, 286-289.

Briselli, M., Mark, E.J. & Dickersin, G.R. (1981) Solitary fibrous tumor of the pleura: eight new cases and review of 360 cases in the literature. *Cancer*, **47**, 2678-2689.

Chan, J.K.C. (1997) Solitary fibrous tumor—everywhere, and a diagnosis in vogue. *Histopathology*, **31**, 568-576.

Dervan, P.A., Tobin, B. & O'Connor, M. (1986) Solitary (localized) fibrous mesothelioma: evidence against mesothelial cell origin. *Histopathology*, **10**, 867-875.

England, D.M., Hochholzer, L. & McCarthy, M.J. (1989) Localized benign and malignant fibrous tumors of the pleura. a clinicopathologic review of 223 cases. *Am. J. Surg. Pathol.*, **13**, 640-658.

Ferreiro, J.A. & Nascimento, A.G. (1996) Solitary fibrous tumor of the major salivary glands. *Histopathology*, **28**, 261-264.

Gangopadhyay, K., Taibah, K., Manohar, M.B. & Kfoury, H. (1996) Solitary fibrous tumor of the parapharyngeal space: a case report and review of the literature. *Ear Nose Throat J.*, **75**, 681-684.

Goodlad, J.R. & Fletcher, C.D. (1991) Solitary fibrous tumour arising at unusual sites: analysis of a series. *Histopathology*, **19**, 515-522.

Guarino, M., Giordano, F., Pallotti, F. & Ponzi, S. (1998) Solitary fibrous tumour of the submandibular gland. *Histopathology*, **32**, 571-573.

Gunhan, O., Yildiz, F.R., Celasun, B., Onder, T. & Finci, R. (1994) Solitary fibrous tumour arising from sublingual gland: report of a case. *J. Laryngol. Otol.*, **108**, 998-1000.

Hanau, C.A. & Miettinen, M. (1995) Solitary fibrous tumor: histological and immunohistochemical spectrum of benign and malignant variants presenting at different sites. *Human Pathol.*, **26**, 440-449.

Havlik, D.M., Farnath, D.A. & Bocklage, T. (2000) Solitary fibrous tumor of the orbit with a t(9 ; 22) (q31 ; p13). *Arch. Pathol. Lab. Med.*, **124**, 756-758.

Kie, J.H., Kim, J.Y., Park, Y.N., Lee, M.K., Yang, W.I. & Park, J.S. (1997) Solitary fibrous tumor of the thyroid. *Histopathology*, **30**, 365-368.

Klemperer, P. & Rabin, C.B. (1931) Primary neoplasms of the pleura. a report of five cases. *Arch. Pathol.*, **11**, 385-412.

Mentzel, T., Bainbridge, T.C. & Katenkamp, D. (1997) Solitary fibrous tumor: clinicopathological, immunohistochemical and ultrastructural analysis of 12 cases arising in soft tissues, nasal cavity and nasopharynx, urinary bladder and prostate. *Virchows Arch.*, **430**, 445-453.

Mohammed, K., Harbourne, G.., Walsh, M. & Royston, D. (2001) Solitary fibrous tumor of the parotid gland. *J. Laryngol. Otol.*, **115**, 831-832.

Morgan, M.B. & Smoller, B.R. (2000) Solitary fibrous tumors are immunophenotypically dis-

tinct from mesothelioma(s). *J. Cutan. Pathol.*, **27**, 451-454.

Rader, A.E., Avery, A., Wait, C.L., McGreevey, L.S., Faigel, D. & Heinrich, M.C. (2001) Fine-needle aspiration biopsy diagnosis of gastrointestinal stromal tumors using morphology, immunocytochemistry, and mutational analysis of c-kit. *Cancer*, **93**, 269-275.

Safneck, J.R., Alguacil-Garcia, A., Dort, J.C. & Phillips, S.M. (1993) Solitary fibrous tumour: report of two new locations in the upper respiratory tract. *J. Laryngol. Otol.*, **107**, 252-256.

Sarlomo-Rikala, M., Kovatich, A.J., Barusevicius, A. & Miettinen, M. (1998) CD117: a sensitive marker for gastrointestinal stromal tumors that is more specific than CD34. *Mod. Pathol.*, **11**, 728-734.

Sato, J., Asakura, K., Yokoyama, Y. & Satoh, M. (1998) Solitary fibrous tumor of the parotid gland extending to the parapharyngeal space. *Eur. Arch. Otorhinolaryngol.*, **255**, 18-21.

Suster, S., Nascimento, A.G., Miettinen, M., Sickel, J.Z. & Moran, C.A. (1995) Solitary fibrous tumors of soft tissue. a clinicopathologic and immunohistochemical study of 12 cases. *Am. J. Surg. Pathol.*, **19**, 1257-1266.

Westra, W.H., Gerald, W.L. & Rosai, J. (1994) Solitary fibrous tumors. consistent CD34 immunoreactivity and occurrence in the orbit. *Am. J. Surg. Pathol.*, **18**, 992-998.

Zukerberg, L.R., Rosenberg, A.E., Randolph, G., Pilch, B.Z. & Goodman, M.L. (1991) Solitary fibrous tumor of the nasal cavity and paranasal sinuses. *Am. J. Surg. Pathol.*, **15**, 126-130.