

Meningeal solitary fibrous tumor: report of a case and literature review

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Folia Neuropathol 2005; 43 (3): 178-185

Abstract

Solitary fibrous tumor is a rare neoplasm that most often involves the pleura. The increasing numbers of this neoplasm have also been reported to date in extrapleural sites.

We report a case of a twenty-four-year-old female with right frontal mass. Histologically, the tumor composed of spindle cell proliferation. Tumor cells were found to be positive for CD34 and CD117 with immunohistochemical studies. Ten months follow-up was uneventful.

Seventy seven cases of meningeal solitary fibrous tumor from the literature are analysed and pathological, immunohistochemical and clinical features are discussed. Solitary fibrous tumor has a slight female predominance, with a male to female ratio of 1:1.5. Age distribution is similar to meningioma ranging from 7-81 years. Approximately 23% of cases originate in the spine which is the most common meningeal location. Histopathologic examination shows uniform spindle cell proliferation with various amount of collagen. CD34-positivity usually allows discrimination from schwannomas, meningiomas and hemangiopericytomas. A differential diagnosis is important because most of the solitary fibrous tumors usually behave in a benign fashion.

In this study, we also showed CD117 (Kit) expression in a case of meningeal SFT. CD117-positivity can be a good strategy for treatment in malignant and recurrent cases. Further investigations are necessary for therapeutic implication of CD117-positivity in SFT.

Key words: *solitary fibrous tumor, meninges, CD34, CD117*

Introduction

Solitary fibrous tumor (SFT), a spindle cell neoplasm, was initially described in the pleura by Klemperer and Rabin in 1931 [27]. Most cases of SFT have been reported to arise in the pleura [7]. They have also been described in numerous non-pleural locations such as the nasal cavity, lung, mediastinum [35], liver, [5] thyroid, [26] cervix, soft tissue [23]. SFTs

have now been recognized at almost all body sites. In 1996, Carnerio et al. reported meningeal SFT as a lesion distinct from fibrous meningioma [11]. Up to date, cases of SFT of the meninges are increasingly being reported. To our knowledge, 77 cases have been reported in the literature.

We presented a case of meningeal SFT that was located in the frontal lobe and reviewed literature. The outcome of the meningeal SFTs reported to date has

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been favourable. However, the biological behaviour is largely unknown. The clinical, histopathological features and behaviour of SFTs are discussed.

Case report

A 24-year-old woman presented to our hospital with one year history of headache and a single complex partial seizure two days before. Physical examination including neurological testing was unremarkable except slight left lower extremity weakness on admission. Magnetic resonance imaging (MRI) demonstrated a dural-based 5.5x4x4cm mass in the right frontal lobe. The lesion was well defined and of heterogeneous intensity. There was slight surrounding edema and compression to the right lateral ventricle, basal ganglia, capsula interna (Fig. 1). MRI findings supported a preoperative diagnosis of an anaplastic astrocytoma. During surgery, a well-circumscribed tumor was observed, and the tumor was found to be attached to the dura at the anterior site of the Sylvian fissure. A total excision of the lesion was performed.

Grossly the tumor was well-circumscribed, firm mass with grey-white whorled cut surface. There was no evidence of necrosis or hemorrhage. Microscopic examination of the surgical specimen revealed a spindle cell neoplasm with rare mitotic figures (0-1 mitoses per 10 high power fields (hpf)). Neoplastic cells were embedded in a highly collagenised stroma (Fig. 2A). The tumor cells had rather indistinct cytoplasmic borders. The nuclear chromatin of the tumor cells was finely dispersed without prominent nucleoli. There was no evidence of nuclear atypia. No definite necrosis and hemorrhage were found.

Immunohistochemical studies displayed diffuse and strong positivity for vimentin (Neomarkers, Fremont, CA, 1/100), CD34 (Dako, Denmark, 1/50), and CD117 (Dako, Carpinteria, CA, 1/50) (Figure 2 B, C, D). Neoplastic cells were negative for epithelial membrane antigen (EMA) (Dako, Denmark, 1/50), S-100 (Neomarkers, Fremont, CA, 1/200), glial fibrillary acidic protein (GFAP) (Neomarkers, Fremont, CA, ready to use), FVIII (Dako, Denmark, 1/25), pancytokeratin (Neomarkers, Fremont, CA, 1/50).

After 10-months of follow-up the patient is asymptomatic and free of the disease.

Discussion

SFTs are rare neoplasms of adult life the vast majority of which arise in the pleura [7,19]. SFTs

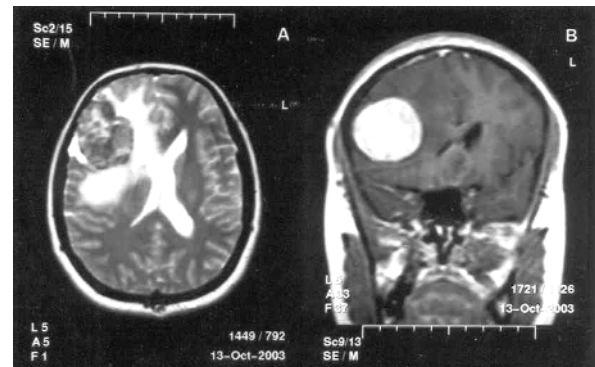


Fig. 1. MRI appearance of dural-based right frontal lobe mass in coronal (A) and axial (B) sections

occur at various extra pleural sites such as the nasal cavity, lung, mediastinum [35], liver [5], thyroid [26], cervix, soft tissue [23]. From Carnerio et al. [11] 77 meningeal SFTs have been reported (Table I).

This review study included 78 cases of SFTs but only 75 cases were accessible for clinicopathological features. This tumor has a slight female predominance, with a male to female ratio of 1: 1.5. Fifty-seven of 75 cases were ranging in age from 7 to 73 years (median 49) and the rest of 18 cases reported by Tihan et al. [50] were in similar age distribution from 7 to 81 years (median 52.5). The reported SFT cases range in size of 1.2 to 9 cm in largest diameter [9,11].

Caroli et al. [12] reported that the posterior fossa and spine are the most common locations for SFTs. Our literature review revealed that SFTs show a tendency to arise in the spine. Seventeen of these tumors were in a spinal location. [3,4,11,12,18,23-25,28,42,50]. Of these 11 tumors were accessible for their locations including cervical (4 cases) [24,25,28,34], thoracic (2 cases) [4,31], lumbar (2 cases) [11,18], cervicothoracic (1 case) [12], thoracolumbar (1 case) [9], and cauda equina level (1 case) [3]. The ratio of spinal to intracranial SFTs was 17: 58. Five cases were located within the ventricular system. Two tumors were in the lateral ventricle [50], two were in the fourth ventricle [29,54] and one was in the third ventricle [30]. Although SFT can arise in any site of the central nervous system such as the basal ganglia, Gasser's ganglion, optic nerve, foramen magnum, there are limited cases in these locations (Table II) [12,28,29].

Histogenesis of SFTs is still unknown. The cell of origin of SFTs can not be defined, but ultrastructural

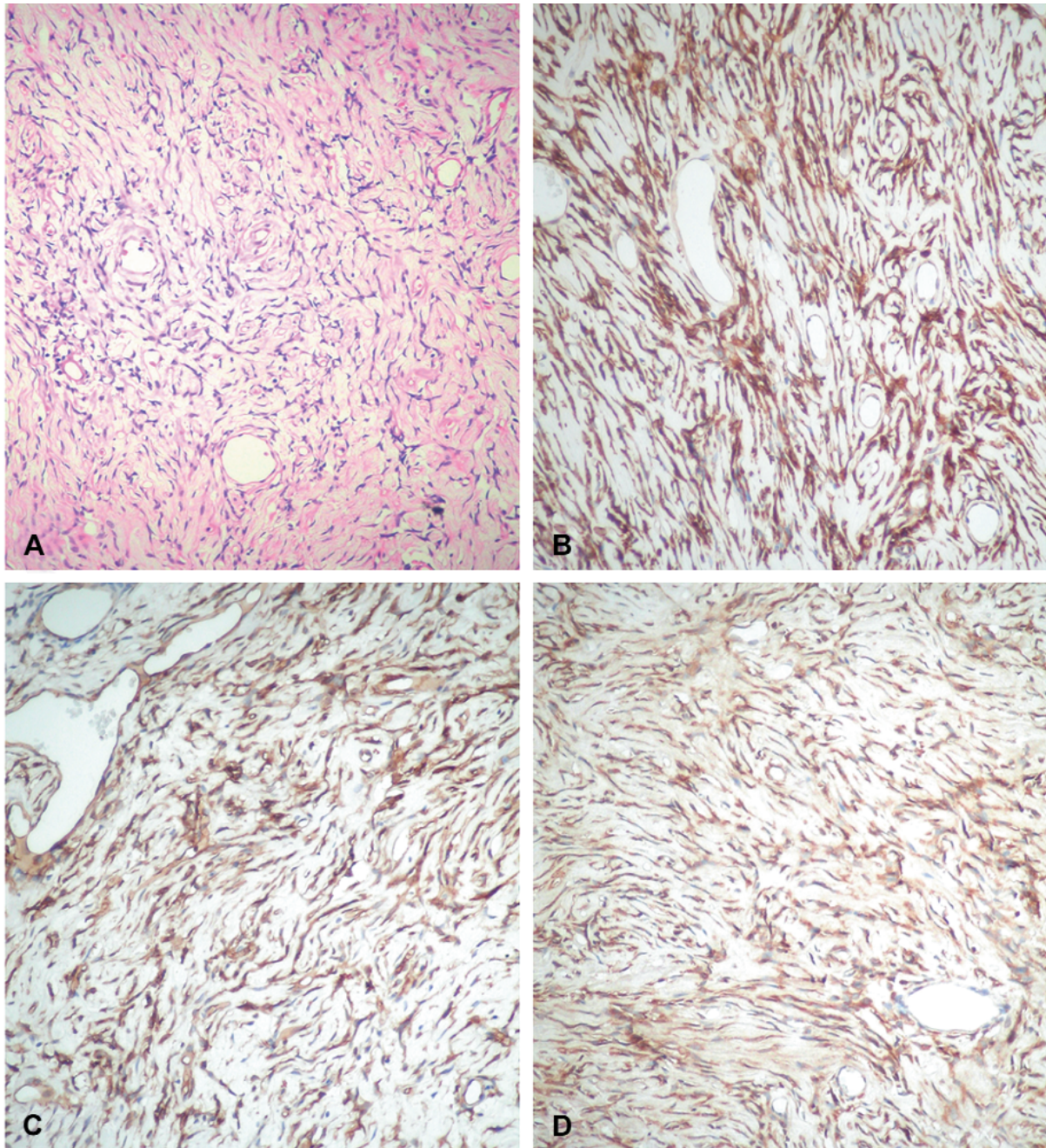


Fig. 2. Light microscopy showed uniform spindle cell proliferation (A) Positive immunoreactivity for vimentin (B), CD34 (C), CD117 (D)

studies favored mesothelial and fibroblastic origin for pleural SFT [7]. Goodlad et al. showed that tumor cells have histological and immunohistochemical features in keeping with fibroblasts and myofibroblasts [22]. Cummings et al. showed CD34 reactivity of dural fibroblasts especially located in the meningeal portion of the dura rather than the periosteal region.

In the same study, CD34 immunoreactivity was identified in the arachnoid or pia mater [17].

SFT occurring at extra pleural sites is sometimes difficult to diagnose because of its histological variability [35,49]. Histological examination usually discloses spindle cell proliferation in a storiform fashion in these tumors. The histopathological

Table I. Review of the clinical features of the solitary fibrous tumor cases

Author/reference	Number of cases	Gender (F:M)	Age	Location (number of cases)
Carnerio et al. [11]	7	5:2	73 51 62 47-63 50-54	Frontal (1) Posterior fossa (1) Tentorial (1) Cerebellopontin angle (2) Spinal (L1-L3and NS) (2)
Perry et al. [42]	3 new cases	3:4	47-73 (54)	Intracranial (NS) (6) Spinal (NS) (1)
Prayson et al. [43]	1	0:1	43	Frontal (1)
Malek et al. [31]	1	0:1	33	Spinal (T7-T8) (1)
Alston et al. [4]	1	0:1	47	Spinal (T4-T5) (1)
Slavik et al. [47]	1	0:1	11	Occipital (1)
Kuchelmeister et al. [28]	2	1:1	64 73	Spinal (C5) (1) Optic nerve (1)
Challa et al. [16]	1	0:1	42	Suprasellar cystern (1)
Zamecnik et al. [54]	1	0:1	45	Intraventricular (4. ventricle) (1)
Nikas et al. [40]	1	0:1	44	Parietal (1)
Brunori et al. [9]	2	1:1	18 46	Spinal (T12-L1) (1) Occipital (1)
Brunnemann et al. [8]	1	0:1	30	Frontal (1)
Gentil Perret et al. [20]	1	1:0	64	Frontal (1)
Kanahara et al. [24]	1	0:1	62	Spinal (C6-C7) (1)
Kataoka et al. [25]	1	1:0	46	Spinal (C4-C5) (1)
Hasegawa et al. [23]	1	1:0	39	Spinal (NS) (1)
Rodriguez et al. [44]	1	0:1	14	Parietal (1)
Nawashiro et al. [37,38]	1	1:0	58	Posterior fossa (1)
Ng et al. [39]	1	1:0	55	Posterior fossa (1)
Suzuki et al. [49]	2	2:0	51 55	Parietooccipital (1) Torcula (1)
Vorster et al. [52]	1	1:0	60	Frontal (1)
Shimizu et al. [46]	1	1:0	50	Falx cerebri (1)
Mordani et al. [34]	1	0:1	33	Spinal (C5) (1)
Morimitsu et al. [35]	2	1:1	59 60	Temporal (1) Frontal (1)
Ahn et al. [2]	1	0:1	57	Temporal (1)
Martin et al. [32]	4	2:2	46 43 72 71	Posterior fossa (1) Cerebellopontin angle (1) Middle fossa (1) Frontoparietal (1)
Barron et al. [6]	1	1:0	61	Frontal (1)
Sanno et al. [45]	1	1:0	29	Posterior fossa (1)
Castilla et al. [14]	1	1:0	40	Intracranial (1)
Gonzalez et al. [21]	1	1:0	51	Occipital (1)
Alameda et al. [3]	1	1:0	35	Spinal (cauda equina) (1)

Table I. Continuation

Donnelan et al. [18]	1	0:1	39	Spinal (L1) (1)
Centeno et al. [15]	1	1:0	25	Occipital (1)
Tihan et al. [50]	18	14:4	7-81 (median 52.5)	Frontal (4) Spinal (NS) (4) Parietal (3) Cerebellar (3) Intraventricular (lateral ventricle) (2) Temporal (1) Occipital (1)
Cassarino et al. [13]	1	1:0	54	Pituitary fossa (1)
Kocak et al. [30]	1	0:1	63	Intraventricular (third ventricle) (1)
Ogawa et al. [41]	1	1:0	69	Tentorial (1)
Kim et al. [29]	4	2:2	7 30 49 55	Basal ganglia (1) Intraventricular (fourth ventricle) (1) Foramen magnum (1) Posterior fossa (1)
Caroli et al. [12]	4	1:3	54 38 29 34	Cerebellar (1) Frontal (1) Gasser's ganglion (1) Spinal (C7-T1) (1)
Our case	1	1:0	24	Frontal (1)

NS, not specified

differential diagnosis of SFT includes other spindle cell neoplasms such as fibrous meningioma [17], meningeal hemangiopericytoma, neurofibroma and schwannoma [11,30]. The distinction between hemangiopericytoma and SFT can be difficult since both of them can have similar histological patterns because of hemangiopericytomatous blood vessels [30]. Especially large lesions had areas of prominent vascularity consisting of thin-walled vessels within the tumor [7]. Fibrous meningioma and hemangiopericytoma mimic the SFT histologically and radiologically [43]. The most important entity in the differential diagnosis is fibrous meningioma. Meningiomas usually show whorls and psammoma bodies [40]. The immunohistochemical findings were helpful for the differential diagnosis. SFTs have been reported to be negative for EMA, S-100, smooth muscle actin, desmin and immunoreactive to CD34, bcl-2 and vimentin [17,49]. The majority of fibrous meningiomas are positive for EMA, some are positive for S-100, while SFT is negative for both markers [40]. CD34 antigen originally described as a marker for human hematopoietic stem cells is likely to be the most useful marker for SFTs [51]. However, immunostaining for CD34 has a limited

value in some cases, CD34 expression is patchy and weaker in hemangiopericytoma where it is rarely observed in fibrous meningiomas [20,36]. Immunoreactivity for CD34 in SFTs is characteristically strong and diffuse [42]. Vimentin is almost always positive in SFT, but this finding is not for a diagnostic value because most of the mesenchymal neoplasms express vimentin [51]. In this case study immunohistochemical findings did not support a diagnosis of schwannian, meningeal or neuroglial neoplasm. These findings are in line with the immunohistochemical features of SFTs.

The clinical behavior of these tumors is unpredictable because of insufficient data. Little is known about malignant behavior of meningeal SFTs. The majority of the SFTs behaved in a benign fashion [7]. The incidence of aggressive behavior is variously reported, as between 13% and 23% of cases in most large series of pleural tumors [7,19]. The published data suggest an indolent behavior for most meningeal SFTs. Three of 78 meningeal SFT metastasized to other sites with a predilection for lung (3/3 lung metastasis) [29,39,41]. Kim et al. [29] reported a case with multiple metastasis including the liver, lung, joint and vertebra. Two of the three

metastatic tumors were located in the posterior fossa and one in the tentorium [29,39,41]. One of 78 reported cases had malignant histologic features, [6] and one of them was widely invasive [14]. Seven of the 73 benign cases have been recurred [11,23,29,37,38,50]. Recurrence was most likely due to the incomplete removal of the tumor [11, 23, 50]. The malignant histological features described for pleural SFTs could be applied for meningeal SFTs. No single histological feature appears to be associated with prognosis [7]. High cellularity, high mitotic count ($\geq 4/10$ hpf) and nuclear pleomorphism seem to be the most useful criteria to predict the malignant behavior [19,43]. In addition, aggressive behavior seems to be in a higher percentage of posterior fossa tumors (2/6 cases) [29,39].

The incidence of SFT among meningeal tumors is relatively low [9]. In the past SFTs were over diagnosed as fibrous meningioma and hemangiopericytoma. Careful histological and immunohistochemical examinations are necessary not to overlook SFT among other meningeal neoplasms [49].

All reported cases underwent surgery as the initial treatment. SFTs seemed to be treated successfully by surgery in most cases. Patients with sub totally resected tumors received radiotherapy [50]. In our case study, the tumor was totally resected and no additional treatment was given.

Kit (CD117) the product of the proto-oncogene *c-kit*, is a tyrosine kinase transmembrane receptor. Kit immunoreactivity is a defining feature of gastrointestinal stromal tumor (GIST) and immunoreactivity for Kit in GIST has been well established [33]. However, data on Kit immunostaining in SFTs is limited. While a few reports describe limited Kit immunoreactivity in pleural SFTs, the proportion of positive cases varies considerably, in small series [1,10]. In the study by Adley et al. [1] the Kit-positivity was reported in 31% pleural SFTs. A study by Butnor et al. [10] revealed expression of Kit in 50% of pleural SFT which was higher than the percentage reported in the previous studies. To our knowledge, the only published study regarding the evaluation of Kit expression in meningeal SFT is that of Kim et al. [29]. They showed only weak Kit staining in one case. We observed diffuse and strong Kit-immunoreactivity in the presented case of meningeal SFT. The success of imatinib in the treatment of GIST has generated interest in

Table II. Location of the solitary fibrous tumor cases

Location	Number of cases	%
spinal	17	22.6
frontal	13	17.3
posterior fossa	6	8.0
parietal	5	6.7
occipital	5	6.7
intraventricular	5	6.7
cerebellar	4	5.3
cerebellopontin angle	3	4.0
temporal	3	4.0
tentorial	2	2.7
others*	12	16.0
total	75	100

*tumors which were less than two cases in any location classified as others

understanding the functional significance of Kit expression in other tumors. SFTs share histological and some immunohistochemical similarities with GIST, including CD34 positivity [10]. Whereas GISTs are consistently immunoreactive for Kit [33], variable results are reported in SFTs [1,10]. The mechanisms underlying Kit expression in SFTs and the relation between GISTs and SFTs are unknown [10]. Further investigations are necessary to evaluate the mechanism of Kit expression and its therapeutic implication in SFTs. Kit immunostaining in SFTs suggests that these tumors are likely to respond to tyrosine kinase inhibitors. In malignant or subtotally resected SFT cases imatinib therapy can be useful.

This review confirms that a majority of SFTs have a benign biological behavior. In general, the reported cases have followed an indolent course. However, experience with these neoplasms is limited [11,23,29,50]. A growing number of malignant SFTs are now recognized. It is advisable to have 10 years or more follow-up.

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