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ORIGINAL ARTICLE

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Solitary fibrous tumors of the meninges

Report of four cases and literature review

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Abstract Central nervous system solitary fibrous tumors are a new pathological entity. To our knowledge, only 60 meningeal solitary fibrous tumors both in the spinal cord and in the brain have been described in the literature. The 56 previously reported cases of meningeal solitary fibrous tumors are critically reviewed. In addition, we report four new cases of solitary fibrous tumors of the meninges. There is a slight male prepoderance. Meningeal solitary fibrous tumors show a tendency to arise in the posterior fossa (26%) and spine (25%). The treatment was mainly total surgical excision. Radiotherapy was given only to four patients with tumors involving the cerebral parenchyma. Sporadic cases of recurrence and distant metastasis have been reported. The prognosis of meningeal solitary fibrous tumors is still unknown because the follow-up of the reported cases is short. It is probable that cases of solitary fibrous tumors of the meninges have been misdiagnosed as other tumors in the past. The best management of these tumors seems to be total surgical excision whenever possible. It is important that every new case of meningeal SFT be reported to throw light on this particular tumor and to affirm its status as a clinicopathological entity.

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Introduction

Solitary fibrous tumor (SFT) is an uncommon spindle cell tumor that typically arises in the visceral pleura [1–4]. Extrapleural sites have been reported, including the pericardium, peritoneum, lung, liver, upper respiratory tract, tunica vaginalis testis, mediastinum, nasal cavities, thyroid and parotid glands, orbit and meninges [3, 5–13]. Histologically, SFTs consist of monomorphic spindle cells organized both into straight, curving or undulating fascicles and unstructured arrangement. Focally prominent bands of hyalinized collagen are characteristic, and the spindle cells are embedded in a conspicuous fibrous matrix [14]. The hystopathogenesis of these tumors has been a matter of debate, and both mesothelial and mesenchymal origin have been proposed [1, 15–20]. Recent studies confirm the mesenchymal origin of SFT [14]. To our knowledge, only 60 cases of SFT of the meninges have been described in the pertinent literature [14, 19, 21–48]. In this report, we describe four new cases of meningeal SFT that fulfill the diagnostic criteria for SFT and critically review the pertinent literature.

Material and methods

Between 1995 and 2000, four cases of SFT of the meninges were surgically treated at our institutes. All tumors were histologically reviewed. The diagnostic criteria for SFT were identical to those used for soft tissues and required the presence of strong CD34 immunoreactivity. The present cases had similar histology and immunohistochemistry. Macroscopically, the tumors were well-circumscribed, tan, and their surfaces were smooth. Upon sectioning, they had a thick, fibrous capsule. Microscopically, the tumor in all cases consisted of spindle cells in a collagenous background. The cellularity and the amount of collagen deposition were variable. Areas of relative hypocellularity with abundant collagen deposition were alternated with more cellular areas. Spindle cells

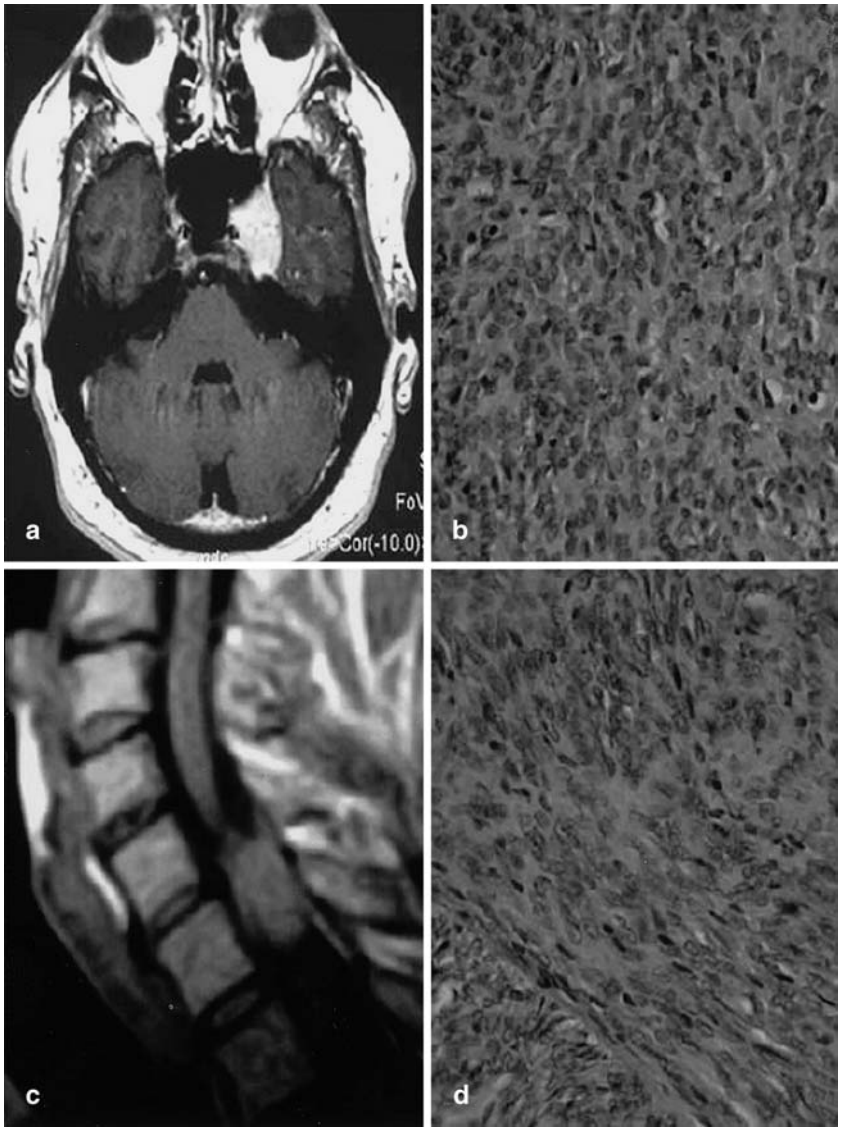

Fig. 1 **a** Case 1: axial contrast-enhanced T1-weighted images showing homogeneous enhancement of a left gasserian tumor. **b** Case 1: photomicrograph showing varying numbers of spindle cells arranged randomly in a collagenous background (H&E). **c** Case 2: sagittal contrast-enhanced T1-weighted images showing dyshomogeneous enhancement of the lesion similar to a neurinoma. **d** Case 2: photomicrograph showing a highly cellular tumor composed of spindle cells arranged in fascicles between bands of collagen (H&E)

were arranged in fascicles in some areas and in an unstructured pattern in others. There were many vessels. There was no evidence of necrosis, hemorrhage or microcystic degeneration. In all cases, MIB-1 labeling was <1%. Immunohistochemical analyses showed strong diffuse reactivity for CD34 and vimentin, but negative reaction for epithelial membrane antigen, S-100 protein and desmin.

Case 1

A 38-year-old man was admitted to our clinic with a 2-year history of right complete trigeminal neuralgia. Magnetic resonance imaging (MRI) demonstrated a right gasserian ganglion lesion with homogeneous enhancement after injection of gadolinium-DTPA (Fig. 1a).

A subtotal removal of the tumor was performed. The patient had a good recovery. The histopathologic diagnosis of the lesion was SFT (Fig. 1b). The postoperative course was uneventful. Fifty months after surgery, the illness was stable.

Case 2

A 54-year-old man presented with a 3-month history of left arm paresthesia and brachialgia. MRI revealed an intraspinal mass at the level of C7-D1 (Fig. 1c). The tumor resembled a spinal neurinoma and showed inhomogeneous contrast enhancement. Total excision was carried out, and the patient had good recovery. The histologic diagnosis of the lesion was SFT (Fig. 1d). There was no clinical or MRI evidence of recurrence after 15 months.

Case 3

A 29-year-old woman presented with a 3-month history of increasing headache, nausea and vomiting. MRI demonstrated a rounded lesion in the right cerebellar hemisphere with tentorial attachment and a small supratentorial extension. The adjacent ventricular recess appeared to be compressed. At operation, a well-demarcated lesion arising from the dura was subtotally excised. A radiotherapeutic course of radiosurgery with LINAC was performed (20 Gy) on the residual disease. The histopathological diagnosis was SFT. The postoperative course was uneventful. The patient was lost to follow-up, but 3 years after surgery was disease-free.

Case 4

A 34-year-old man presented with a 1-year history of headache and left limb weakness. Neurologic examination showed a slight left hemiparesis (4/5). MRI revealed a mass located in the right frontal lobe. The lesion did not enhance after injection of gadolinium-DTPA. The tumor was totally excised. The postoperative course was uneventful, and hemiparesis resolved completely. A diagnosis of SFT was made. Five years after surgery, the patient remains well.

Discussion

Most cases of SFT have been reported to arise in the visceral pleura [1–4]. However, these tumors have now been documented in many other sites outside the thoracic cavity [3, 5–13]. SFT of the meninges was first reported as a lesion distinct from fibrous meningioma by Carneiro et al. in 1996 [24]. Since then, a further 53 cases of SFT have been reported directly involving the neuraxis, both in the spinal cord and in the brain [14, 19, 21–48]. The reports of meningeal SFT accessible to us include 45 cases in the brain and 15 in the spine. Among 18 cases reported by Tihan et al. [44], four non-meningeal cases are included, and three cases reported by Perry [39] are partially described, so that, including the present series, we have found that meningeal SFT occurred in 20 women and 23 men with a mean age of 47.6 years (range: 11 to 73 years; Tables 1, 2). Meningeal SFTs show a tendency to arise in the posterior fossa (26%) and spine (25%). SFTs of the central nervous system typically show a clear dural origin so that they may mimic meningiomas radiographically and surgically [34]. However, most spinal SFTs have no dural attachment (Table 2).

The histopathogenesis of SFTs has been debated, but now the mesenchymal origin of these tumors is generally accepted on the base of immunohistochemical, ultrastructural and cell culture evidence [6, 14, 49]. Immunohistochemical studies of SFT, showing strong CD34 reactivity of the cells, suggest that this tumor is mesenchymal in nature. CD34 is a transmembrane glycoprotein that has been found in hematopoietic stem cells, endothelial cells and an ubiquitous sub-population of fibroblasts, from which SFT may originate [50–54]. Furthermore, negative immunoreactivity to epithelial markers and, ultrastructurally, the absence of mesothelial structures confirm a mesenchymal origin of these tumors. Therefore, SFT should be classified into the group of rare

Table 1 Intracranial fibrous solitary tumor. S*, extension of surgery unknown; RT, radiotherapy; b.i., brain invasion; R, recurrence M, metastasis; n.s., not stated

Reference	Age/sex	Location	Removal	Postoperative course	Follow-up
[24]	51/F	Posterior fossa	Total	No	Alive, 20 years
	47/M	Cerebellopontine angle	Total+RT (b.i.)	No	Alive, 10 years
	73/M	Frontal lobe	Total	No	Alive, 8 months
	62/F	Tentorium	Total	No	Alive, 7 months
	63/F	Cerebellopontine angle	Total	No	Alive, 9 months
[39]	3 new cases	n.s	n.s.	n.s.	n.s.
[40]	43/M	Frontal lobe	Total	n.s.	n.s.
[32]	73/F	Optic nerve	Total	n.s.	n.s.
[26]	42/M	Tentorium	n.s.	No	Alive, 1 year
[14]	18/M	Craniospinal	Total	No	Alive, 12 months
42	11/M	Occipital	Total	n.s.	n.s.
43	45/M	Meningeal (region of the 4th ventricle)	S*	No	Alive, 15 years
[23]	30/M	Frontal	S*+RT (b.i.)	No	Alive, 9 months
[28]	64/F	Falcine (diagnosis meningioma)	Total	1 R: 30 years	Died postoperatively
[38]	44/M	Parietal parasagittal	Subtotal	n.s.	n.s.
[41]	14/F	Parietal convexity	Total	n.s.	n.s.
[36]	58/F	Posterior fossa	Total	No	n.s.
[37]	55/F	Posterior fossa	Total+RT	3 R over 8 years, M: 9 years (lung, neck)	Alive, 10 years
[48]	60/F	Frontal	n.s.	No	Alive, 18 months
[47]	51/F	Transverse sinus	Subtotal	1 R: 15 years	Alive, 7 years
	54/F	Parieto-occipital	Total	No	Alive, 3 years
[45]	61/F	Deep frontal	Near-complete+RT	No	Alive, 10 months
[34]	46/F	Posterior fossa	Total	No	Alive, 3 years
	43/M	Cerebellopontine angle	Almost complete	No	Alive, 2 years
	72/F	Middle fossa	Subtotal	No	Alive, 1 years
	71/M	Frontoparietal	Incomplete	–	Died, postoperatively
[21]	58/M	Temporal lobe	Total	No	Alive, 1 year
[25]	25/F	l occipital	Total	No	Alive, 3 years
[44]	12 cases	9 supratentorial, 3 cerebellar	–	–	–
This study	38/M	Gasser's ganglion	Total	No	Alive, 15 months
	29/F	Cerebellar	Total	No	Alive, 3 years
	34/M	Frontal	Total	No	Alive, 5.5 years

Table 2 Spinal solitary fibrous tumor. S*, extension of surgery unknown; im, intramedullary; id, intradural; ed, extradural; em,extramedullary; td, transdural; im, intramedullary; R, recurrence; n.s., not stated

Reference	Age/sex	Location	Dural origin	Removal	Recurrence	Follow-up
[24]	50/F	Spinal cord	No	Subtotal	R: 5 years	Died, 5 years
	54/F	L1-L3	Yes	Total	No	Died, 7 years
[32]	64/M	C5 (id-im)	No	Total	n.s.	n.s.
[33]	33/M	T7/T8 (id-em)	No	Total	No	n.s.
[22	47/M	T4-T5 (id-em)	No	Total	No	Alive, 2 months
[30]	62/M	C6-C7 (ed-im)	No	n.s.	No	n.s.
[31]	46/F	C4-C5 (td-em involving C5 rootlets)	Yes	S*	n.s.	n.s.
[29]	39/M	n.s.	Yes	S*	2 R: 5 and 9 years	Alive, 9 years
[14]	46/F	T12-L1 (td-em)	No	Total	No	Alive, 4 months
[27]	39/M	L1 (ed)	No	Total	No	n.s. (“long-term follow-up”)
[49]	51/M	T2-T3 (id-em)	No	Total	No	Alive, 7 months
[35]	33/M	C5 (im)	No	Total	No	Alive, 18 months
[44]	2 cases	–	–	–	–	–
This study	54/M	C7/D1 (id-em)	No	Total	No	Alive, 15 months

mesenchymal, non-meningothelial tumors of the central nervous system. SFTs are usually negative also for vascular, neural crest and muscle markers [15, 34, 54] and are positive for vimentin. It should be highlighted that CD34 reactivity is not an exclusive marker of SFTs, but it can be identified also in meningiomas, neurofibromas and hemangiopericytoma as a result of the inclusion of dural fibroblasts within tumors. However, the pattern of CD34 reactivity in this latter tumor is weak and patchy, [34, 39, 51]. Immunoreactivity for bcl-2 has often been identified in SFTs and may be useful in the differential diagnosis [9, 22, 29, 34, 39, 51]. SFTs show distinctive cytological features that allow diagnosis in cytological specimens [55]. Clayton et al. [55], in a recent study, reported as characteristic features of SFT: the presence of oval to polygonal tumor cells, cellularity ranging from scant to moderate, a background containing irregular ropy elements of collagen and a few inflammatory cells, loose aggregates of cells enmeshed in a collagen matrix and nuclei uniformly bland, with evenly distributed, fine granules of chromatin. SFTs can mimic other tumors, rendering diagnosis difficult [4]. This is especially true when SFTs arise in uncommon sites such as the neuraxis. It is fundamental to be aware of this tumor and know that it must be included in the differential diagnosis with hemangiopericytoma and fibrous meningioma, schwannoma, neurofibroma and fibrosarcoma [24, 38, 39, 51]. This distinction is imperative, especially for those tumors that require postoperative radiotherapy. The fact that SFT has been diagnosed with increasing frequency in recent years as the result of improved methods of pathologic examination is reassuring.

The natural history of SFT is not completely defined because the period of follow-up of the reported cases of SFT is quite short. Histologically, the majority of SFT appear to be benign, but an analysis of 223 pleural SFT identified 82 cases with atypical features [4]. Clinically, 13–23% of pleural SFTs manifest with local invasion, recurrence, intrathoracic spread or distant metastasis [51]. SFT of the meninges generally pursues a slow, indolent and non-aggressive course, with only rare recurrence or metastasis [2, 28, 29, 37]. The malignant histological features described for SFTs of the pleura could be applied for meningeal SFTs. These malignant features include high cellularity, a high number of mitotic figures, necrosis and nuclear pleomorphism [4]. Three examples of clinically and histologically malignant meningeal SFTs have been reported [4, 29, 44]. All these cases showed an increased number of mitotic figures, hypercellularity, high proliferation index and nuclear pleomorphism. However, it has been noted that if the tumor is amenable to total removal, the above-mentioned histological findings are not predictive of malignant behavior [1, 3, 50]. Usually, the glucose metabolic rate of tumors is a good predictor of their malignancy [26]. Unfortunately, only one case of meningeal SFT studied with FDG-PET has been described [56]. In this case, FDG-PET demonstrated faint uptake of glucose in the tumor, which was of malignant nature. Also, expression of CD34 could be a useful predictor for SFTs because it may be lost focally in some high-grade tumors [44, 57]. Complete removal, rather than histological appearance, seems to be the most important prognostic marker of SFT [1, 21, 34]. This is mainly suggested by the experience with SFT at other sites, because the follow-up period is quite short in most cases of meningeal SFTs reported until now. SFTs seem to be successfully managed by surgery alone. A total excision has been possible for most meningeal SFTs, and evidence of recurrence or metastasis were present in rare cases [23, 24]. The role of postoperative radiotherapy in meningeal SFTs with atypical features is unknown because very few cases have been described. It is interesting to note that two spinal cases recurred and three intracranial cases recurred, of which one metastasized to the cervical lymph nodes and lung parenchyma. Unfortunately, it is difficult to establish if the recurrence rate of these meningeal types is a lot lower than that of the pleural (13–23%) or if this differences results from the shorter follow-up. In conclusion, prognosis of meningeal

SFT remains unclear; consequently, careful and long-term follow-up remain mandatory for all meningeal SFTs.

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