Interdisciplinary Neurosurgery 21 (2020) 100737

Contents lists available at ScienceDirect

Interdisciplinary Neurosurgery

journal homepage: www.elsevier.com/locate/inat

Case Reports & Case Series

Long term recurrence of solitary fibrous tumor involving vertebral body in thoracic spine. A case report

Alessandro Di Bartolomeo[a,b,*], Tanguy Fenouil[b], Marco Giugliano[c], Rostom Messerer[a], Eurico Freitas[a], Cédric Barrey[a]

[a] Spine Surgery Department, Lyon University Hospital, 52 Boulevard Pinel, 69,500, Bron, France
[b] Department of Pathological Anatomy and Cytology, Lyon University Hospital, 59 Boulevard Pinel, 69,394, Bron, France
[c] Department of Neurosurgery, Policlinico Umberto I, University of Rome "La Sapienza". 155 Viale del Policlinico, 00161 Rome, Italy

ARTICLE INFO

Keywords:
Solitary fibrous tumours
Spinal
Local recurrence

ABSTRACT

Solitary fibrous tumor (SFT) represents only 0.08% of all primary bone tumors and 0.1% of primary malignant bone tumors and rarely occurs in the spine. We present the case of a 56-year-old woman with long term recurrence (11 years) of spinal SFT involving T8 vertebral body. We performed a total resection of the lesion and spinal fusion T6-T11 with T8 titanium mesh and placement of pedicle screws in T6-T7 and T10-T11 connected by rods. Microscopic examination confirmed the recurrence of the WHO grade II solitary fibrous tumor. SFT is known for a late but common recurrence and uncertain behaviour. Gold standard treatment is Gross Total Resection. We believe that when vertebral bone is involved it is essential to perform a total excision with "supracomplete" resection if possible in order to avoid local recurrence, more difficult to treat due to an higher rate of perioperative complications. Periodical long-term follow-up is essential to allow early detection of relapses and to allow long-term survival.

1. Introduction

Solitary fibrous tumor (SFT) is a rare fibroblastic mesenchymal tumor of soft tissue, the putative cell of origin was debated [1]; it represents only 0.08% of all primary bone tumors and 0.1% of primary malignant bone tumors and rarely occurs in the spine. Previously classified with hemangiopericytomas, this lesion was first described in 1931 by Klemperer and Rabin [2]. It is described mainly in the pleura, but recent studies have shown that it is a ubiquitous tumor [3,4,5]. The natural history is usually benign even though various cases show an aggressive behaviour. In current literature some cases of malignancy and recurrence are reported [6]. We describe a case of recurrent spinal SFT involving thoracic spine (T8 posterior wall) occurring more than 10 years after primary surgery.

2. Case report

We present the case of a 56-year-old woman treated in 2005 with dorsal decompression and total excision of an extramedullary lesion located left-anterior to spinal cord at the level of T8, turned out to be an SFT upon histological examination. She reported complete resolution of pain after the intervention. No preoperative imaging data was available.

The new beginning of the symptomatology started back in 2016 by the occurrence of back pain located in the region of the previous surgical suture. Back pain was accompanied by intercostal neuralgia in the left T8 hemi-dermatome. Symptoms remained stable for few months. Afterwards, she developed gait disturbance with typical neurogenic claudication and episodes of intense cramps and neuropathic pain in the lower limbs. Successive MRIs demonstrated a local recurrence of the tumor, which appeared as a lesion of $25 \times 9 \times 20$ mm (cranio-caudal × anteroposterior × transverse diameters) extended from the posterior wall of T8 to epidural space, more on the left, determining a compression on the spinal cord (Fig. 1). In relation to imaging showing tumor recurrence and neurological risk of paraplegia, a surgical indication for complete resection was given. Then a posterior vertebrectomy was performed, with radical excision of the lesion, and spinal fusion with T8 titanium mesh and pedicle screws in T6-T7 and T10-T11 connected by rods. Total resolution of symptoms was obtained. 6- and 12-months follow-up showed no recurrence.

3. Surgical technique

Later exposure of posterior spine between T6 and T12 epidural scar

* Corresponding author. Department of Neurosurgery, Policlinico Umberto I, University of Rome "La Sapienza". 155 Viale del Policlinico, 00161 Rome, Italy.
E-mail address: diba.ale@gmail.com (A. Di Bartolomeo).

https://doi.org/10.1016/j.inat.2020.100737
Received 2 March 2020; Received in revised form 25 March 2020; Accepted 29 March 2020

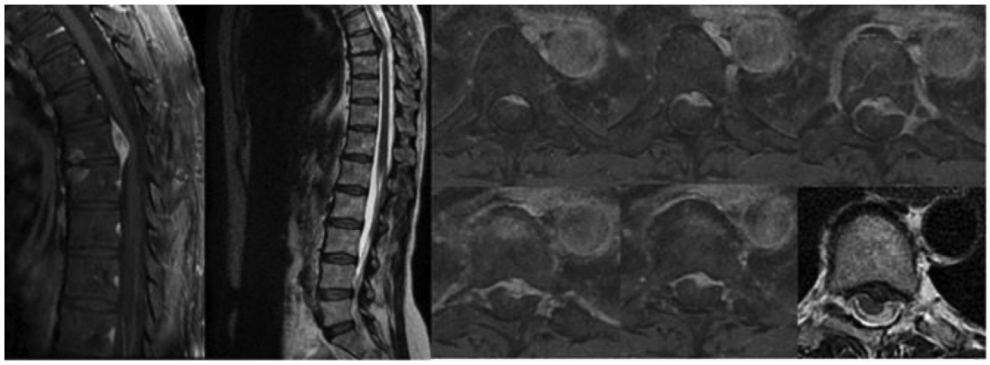

Fig. 1. Lesion on the posterior surface of the T8 body exerting a mass effect on the spinal cord, which extends laterally beyond the whole T8 body and in craniocaudal axis beyond T7-T8/T8-T9 intervertebral discs. Shows intense contrast enhancement with regular margins and central residual hypointensity; insertion appears to be on the whole posterior face of the vertebral body.

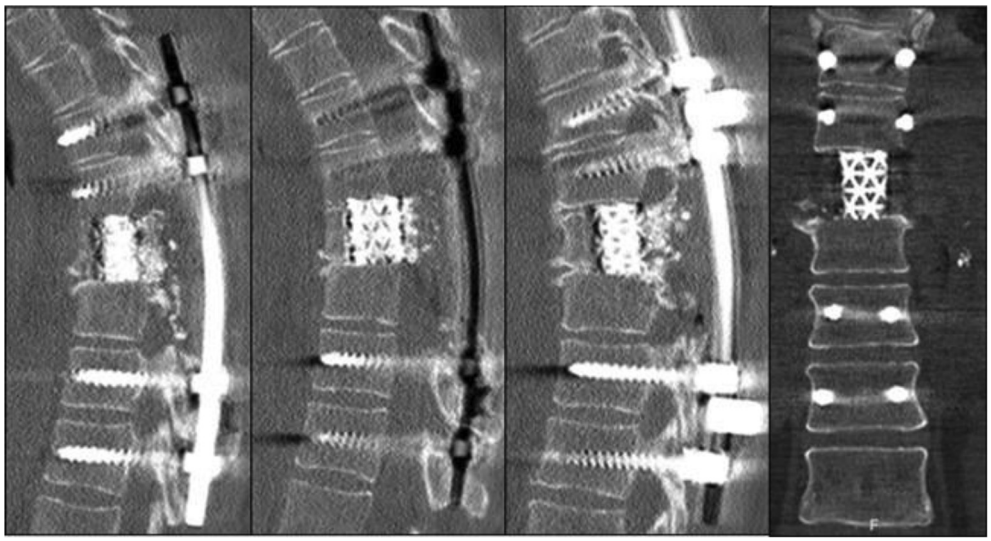

Fig. 2. Posterior T8 hemicorporectomy with osteosynthesis material (somatic cage and graft). Laminectomy T8-T9. Osteosynthesis material (rods and screws) in T6-T7-T10-T11.

tissue was found next to T8 on the left (due to previous intervention with left translaminar approach). A VCR (vertebral column resection) type resection in T8 was performed: we found tumoral tissue perfectly consistent with the data of preoperative imaging in the form of left posterolateral epidural recurrence, so we performed a complete excision of the pathological tissue; the tumor was easily dissected by dura mater but not by T8 posterior wall thus leading us to suppose bone origin, we therefore proceeded to a sub-complete vertebral resection in order to achieve surgical radicality, with complete resection of the residual posterior arch of T8, excision of the two pedicles as well as the vertebral body and the two adjacent disks. Only the anterior cortex of the vertebra (T8) was respected. Mesh reconstruction cage filled with a phosphocalcic bone substitute was placed for anterior intervertebral T7-T9 fusion and additional posterior stabilization with placement of pedicle screws in T6-T7 and T10-T11 connected by rods was performed to avoid pull-out of the mesh. We also used a complementary reinforcement rod on the right side connected between T6-T7 and T10-T11 (Fig. 2). Tissue was sent for anatomopathological examination and histological analysis.

4. Anatomopathological findings

Microscopic examination found a very sparse tumoral proliferation, made of fusiform cells with poorly visible cytoplasm and elongated nuclei, sometimes in ripples evolving within a relatively dense fibrous stroma. The vascularization was made of capillaries with relatively thin walls. The cell density remained moderate, the anisocaryose was not marked, it was not seen mitosis on 10 fields at 40 magnification. The lesion infiltrated the epidural tissue and the elastic fibres. After immunohistochemical technique on fixed sections, we noted:

- Protein S100: negative
- Neurofilament protein: negative
-CD34: positive (membrane expression by endothelial cells and some tumoral cells)
- Proliferation index (Ki-67): not deductible
- Stat 6: positive (nuclear expression by tumoral cells)

In conclusion, the microscopic examination confirmed the recurrence of the WHO grade II solitary fibrous tumor (Fig. 3).

5. Discussion

The most frequent location of SFT is intrathoracic; however, extrathoracic localization is well described in the literature. The origin of the spinal cord SFT can be difficult to determine, and it has been reported to occur: intramedullary (58%), intradural-extramedullary (24%), and extradural (18%). The thoracic spine is predominantly involved (56.3%), followed by the cervical spine (31.2%), with average age of diagnosis of 46,5 years (male to female ratio of 1,4/1) [7]. To the best of our knowledge there are only 8 cases reported of SFT with the involvement of a vertebral body [8,9,10,11,12,13,14,15] (Table 1). SFTs have been previously grouped together with hemangiopericytomas. The pathological features of SFT have recently been reviewed by the WHO in a new classification of soft tissue tumors [16]. Given the most frequent origin of this tumour from pleura, it was initially thought to be of mesothelial derivation. However, the further immunohistochemical characterization and its ubiquitous localization

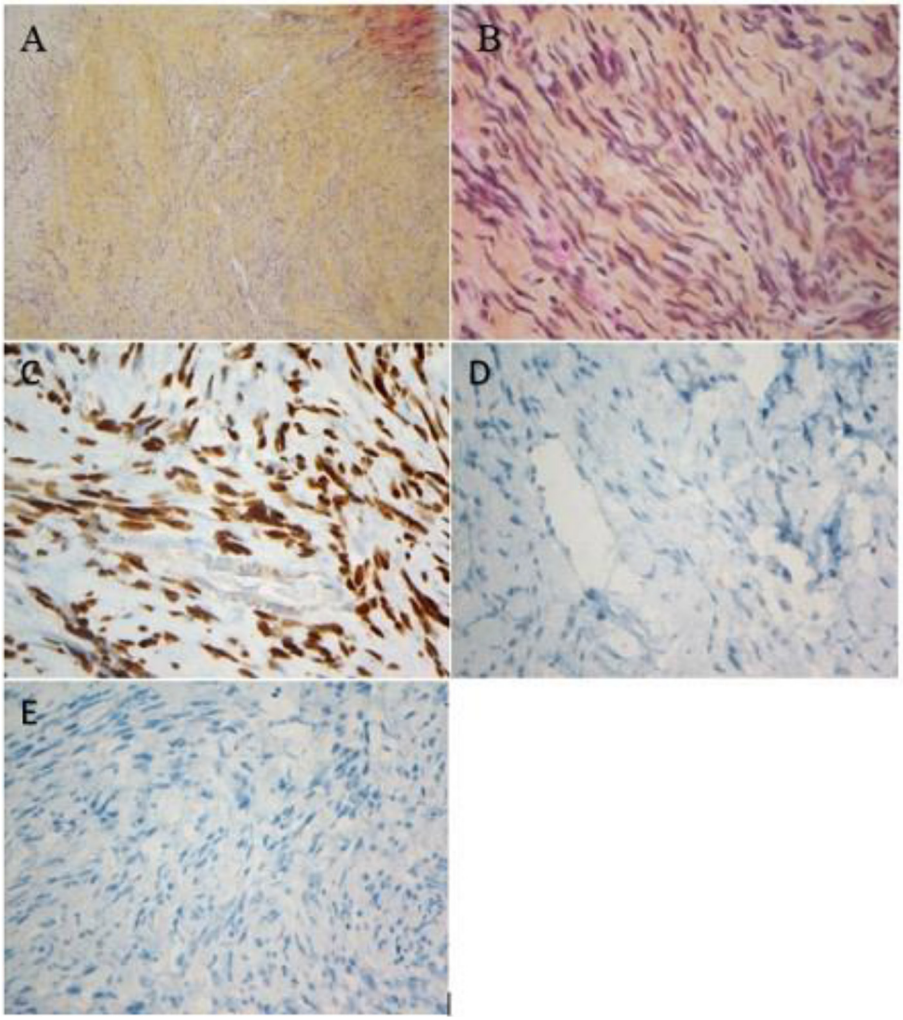

Fig. 3. Microscopic features of the relapsing solitary fibrous tumor/haemangiopericytoma. A) Low magnification (HESx5) where the patternless pattern is seen. It corresponds to intermixed fibrous acellular and more cellular areas associated with staghorn vessels. b) High magnification (HESx40) with high cellular field composed of uniform, bland and fusiform cells. They have scant cytoplasm and elongated to wavy nucleus. There is no mitotic figure or atypia, as well as no hypercellularity corresponding to grade II. c) High magnification (x40) immunochemistry showing a strong nuclear expression of STAT6 protein, characteristic of the solitary fibrous tumor/haemangiopericytoma. Meningiomas do not have nuclear staining for STAT6 protein. d) and e) respectively negative staining of the tumor cells for PS100 and NF that rule the differential diagnostic out (schwannoma and neurofibroma).

allowed to reveal its mesenchymal origin [17]. There are no specific radiological features which distinguish the solitary fibrous tumor. The CT shows a soft tissue, that can cause bone erosion at the level of the vertebral column with homogeneus enhancement. No calcifications are usually identified on CT. In some cases, larger tumors tend to have areas of necrosis especially in the central region represented by hypodense areas on CT. MRI findings of SFT show a hypointense mass expanding into or eroding adjacent bone on T1 and T2-weighted imaging. T1 postcontrast images show vivid, homogenous enhancement of the mass [18,19]. Short T1 inversion recovery imaging would demonstrate osseous invasion as a hyperintense component with the normal marrow being suppressed. However, variable characteristics on MRI have been reported (lesional necrosis or haemorrhage).

The histology is generally comprised of collagenous matrix with arrays of haphazardly arranged, uniform elongated spindle cells [20]. Immunohistochemistry displays positive stain for CD34 in SFTs [21]. With the identification of CD34 and STAT6 in SFTs, the diagnosis has become less vague than it was in the past. In the literature from 10 to 15% showed a malignant behaviour [21]. Factors of unfavourable prognostic significance are: tumor size (greater than5cm), the site involved (thoracic, mediastinal or retroperitoneal), high cellularity, high mitotic index (more than four mitoses for 10 × 400 fields), areas of necrosis, negative CD34 staining. However, tumors without unfavourable prognostic characters can develop a malignant behaviour [22]. Regarding spinal localization, only one case with distant metastasis is reported in the literature [23]. SFT is known for a late but common recurrence. A study aiming to determine outcomes for late recurrence reported a median time of 12 years (first recurrence) [24]. The most common pattern of recurrence is local. Gold standard treatment to avoid local recurrence is the total surgical resection. In case of residual disease (not surgically removable) could be indicated adjuvant radiotherapy. No alternative therapy to surgery to date has proven to be conclusive. Periodical long-term follow-up is essential to allow early detection of relapses and to allow long-term survival [25].

6. Conclusions

Spinal SFT produces non-specific clinical manifestations related to slowly progressive compression of nerve structures. In our case, the patient presented back pain accompanied by intercostal neuralgia and more advanced gait disorders. The time of recurrence was 11 years. In this case radiological features were not clear about bone involving. Intraoperative findings allowed us to identify T8 posterior wall cortex involving and we therefore performed a large bone amputation to avoid a new local recurrence. In conclusion we believe that when vertebral bone is involved it is essential to perform a total excision with “supracomplete” resection if possible in order to avoid local recurrence, because recurrences are common and more difficult to treat surgically than the primary tumor, requiring a more extensive resection and often a spinal arthrodesis with increased rate of perioperative complications [25,26]. Further studies must be carried out to standardize best surgical technique and any adjuvant radiotherapy treatment.

Table 1
Review of literature.

Author	Sex/Age	Level	Treatment	Follow up
Hui-Yuan Su et al. 2019	F/46	T4-T6 intrathoracic extradural lesion involving vertebral bone e compressing spinal cord	Total resection	ND
Mizuo Ando a et al. 2019	M/57	Soft tissues of the neck involving C2-C3 bone and left vertebral artery	Total resection	4 Years
Naoki Oike1 et al.2017	F/49	L1 posterior arch (malignant SFT)	Total resection and chemotherapy	5 Years later with lung metastasis; 8Y later dead for SFT
Zerwa Farooq et al. 2016	M/73	L1 body (recurrence after 10 years)	Total resection	ND
Nao Tsutsumi et al. 2014	M/19	Right neck soft tissues involving arch of C1 and C2 body (recurrence after 10 years)	Total resection	12 months
B. Bouyer et al. 2012	F/56	Extradural involving T10-T11 vertebral bone third recurrence after 4 years	Total resection	12 months
Koji Hashimoto 2007	F/71	Soft tissues of the neck involving C5 body	Total resection	6 months (first recurrence)
Rory B. Donnellan 2000	M/39	L1 posterior wall	Total resection	Long term F.Up uneventful referred

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Bibliography

[1] K. Thway, W. Ng, J. Noujaim, R.L. Jones, C. Fisher, The current status of solitary fibrous tumor: diagnostic features, variants, and genetics, Int J Surg Pathol. 24 (4) (2016 Jun) 281–292 Epub 2016.
[2] Klemperer P, Rabin CB: Primary neoplasms of the pleura. A report of five cases. Arch Pathol 11:385–412, 1931.
[3] C.D. Fletcher, The evolving classification of soft tissue tumours: an update based on the new WHO classification, Histopathology 48 (2006) 3e12.
[4] C. Gengler, L. Guillou, Solitary fibrous tumour and haemangiopericytoma: evolution of a concept, Histopathology 48 (2006) 63e74.
[5] Y. Zhu, K. Du, X. Ye, D. Song, D. Long, Solitary fibrous tumors of pleura and lung: report of twelve cases, Thorac Dis 5 (3) (2013) 310e3.
[6] C.A. Hanau, M. Miettinen, Solitary fibrous tumor: histological and immunohistochemical spectrum of benign and malignant variants presenting at different sites, Human Path 26 (1995) 440e9.
[7] Munoz E, Prat A, Adamo B, Peralta S, Ramon, Cajal S, et al. A rare case of malignant solitary fibrous tumor of the spinal cord. Spine (PhilaPa 1976) 2008;33:E397—9.d.
[8] R.B. Donnellan, D. Govender, S.H. Chite, A.T. Landers, An unusual presentation of solitary fibrous tumor, Spine 25 (2000) 749e51.
[9] K. Hashimoto, K. Miyamoto, H. Hosoe, G. Kawai, K. Kikuike, K. Shimokawa, et al., Solitary fibrous tumor in the cervical spine with destructive vertebral involvement: a case report and review of the literature, Arch Orthop Trauma Surg (2008;128(10):) 1111e6.
[10] B. Bouyer, N. Guedj, G. Lonjon, P. Guigui, Recurrent solitary fibrous tumour of the thoracic spine. A case-report and literature review, Orthop Traumatol Surg Res (2012;98(7):) 850e3.
[11] Farooq Z, Badar Z, Zaccarini D, Tavernier FB, Mohamed A, Mangla R. Recurrent solitary fibrous tumor of lumbar spine with vertebral body involvement: imaging features and differential diagnosis with report of a case. Radiology case reports II (2016) 450–455.
[12] N. Tsutsumi, Y. Kojima, K. Nishida, K. Maeno, K. Kakutani, F. Kawakami, et al., Surgical treatment for recurrent solitary fibrous tumor invading atlas, Head Neck. 36 (11) (2014 Nov) E121–E124.
[13] N. Oike, H. Kawashima, A. Ogose, T. Hotta, T. Hirano, T. Ariizumi, et al., A malignant solitary fibrous tumour arising from the first lumbar vertebra and mimicking an osteosarcoma: a case report, World J Surg Oncol. 15 (1) (2017) 100.
[14] Ando M, Kobayashi H, Shinozaki-Ushiku A, Chikuda H, Matsubayashi Y, Yoshida M et al. Spinal solitary fibrous tumor of the neck: Next-generation sequencing-based analysis of genomic aberrations. Auris Nasus Larynx. 2019 Dec 23. pii: S0385-8146(19)30932–0.
[15] H.Y. Su, T.H. Tsai, S. Yang, J.Y. Lee, Dumbbell-shaped solitary fibrous tumor of thoracic spine, Kaohsiung J Med Sci. 35 (8) (2019 Aug) 517–518.
[16] C. Gengler, L. Guillou, Solitary fibrous tumour and haemangiopericytoma: evolution of a concept, Histopathology 48 (2006) 63–74.
[17] Langman G. Solitary fibrous tumor: a pathological enigma and clinical dilemma. J Thorac Dis 2011;3(2):86e7.
[18] Mariniello G, Napoli M, Russo C, Briganti F, Giamundo A, Maiuri F, et al. MRI features of spinal solitary fibrous tumors. A report of two cases and literature review. Neuroradiol J 2012;25(5):610e6.
[19] D.T. Ginat, A. Bokhari, S. Bhatti, Dogra V. Imaging features of solitary fibrous tumors, Am J Roentgenol 196 (2011) 487e95.
[20] T. Mentzel, T.C. Bainbridge, D. Katenkamp, 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 (1997) 445e53.
[21] A.V. Vallat-Decouvelaere, S.M. Dry, C.D. Fletcher, Atypical and malignant solitary fibrous tumors in extrathoracic locations: evidence of their comparability to intrathoracic tumors, Am J Surg Pathol 22 (1998) 1501–1511.
[22] M. De Perrot, A.M. Kurt, J.H. Robert, B. Borisch, A. Spiliopoulos, Clinical behavior of solitary fibrous tumors of the pleura, Ann Thorac Surg 67 (1999) 1456–1459.
[23] Munoz E, Prat A, Adamo B, Peralta S, Ramon, Cajal S, et al. A rare case of malignant solitary fibrous tumor of the spinal cord. Spine (PhilaPa 1976) 2008;33:E397—9.
[24] F.D. Beaman, M.J. Kransdorf, D.M. Menke, Schwannoma: radiologic-pathologic correlation, Radiographics (2004;24(5):) 1477e81.
[25] L.A. Robinson, Solitary fibrous tumor of the pleura, Cancer Control 13 (2006) 264–269.
[26] Qi Jia Zhenhua, Zhou Dan, Zhang Jian, Yang Chao, Liu Ting, Wang Zhipeng Wu et Al. Surgical management of spinal solitary fibrous tumor/hemangiopericytoma: a case series of 20 patients. Eur Spine J. 2017 Nov 10:1–11.