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Calcifying pseudo-tumor of the spine: description of a case and review of the literature

Received: 3 September 2002
Revised: 8 February 2003
Accepted: 8 February 2003
Published online: 21 May 2003
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Abstract A case of calcifying pseudo-tumor of the thoracic spine, a rare lesion with tumor-like behavior and a probable inflammatory-reactive origin, is described. The clinical-pathological and neuro-radiological aspects of this lesion are discussed in relation to surgical treatment. In accordance with the other cases reported in the literature, the case observed confirmed the benign behavior of the lesion and the effectiveness of surgical treatment for achieving complete resolution of clinical symptoms without any recurrences, even when removal is only subtotal.

Keywords Pseudo-tumor · Fibro-osseous lesion · Spine

Introduction

Calcifying pseudo-tumor is a rare lesion, of which only a few cases localized in the spine have been described [1, 2, 3, 4, 5]. The most important case series is the one reported by Bertoni [1] and co-workers in 1990, comprising 14 cases, of which 8 were situated in the spine. In the present paper, we describe a case of calcifying pseudo-tumor of the thoracic spine.

Case report

A 40-year-old man was referred to us with a 2-month history of thoracic pain radiating to the anterior surface of the chest, accompanied by progressive difficulty in walking. Neurological evaluation revealed a spastic paraparesis and a bilateral level of hypoaesthesia at T8-T9. Results of routine laboratory tests were normal. Magnetic resonance imaging (MRI) of the thoracic spine documented an extradural lesion at T8-T9, occupying the lateral recess and the right conjugate foramen and extending cranio-caudally; this lesion was hypointense on both T1- and T2-weighted images and did not present enhancement after gadolinium administration

(Fig. 1, Fig. 2). Contrast-enhanced computed tomography (CT) demonstrated, at the same site, a hyperdense, non-homogeneous lesion, compatible with calcified tissue (Fig. 3). At surgery, via a T8-T9 laminectomy, a nodular, encapsulated lesion with a maximum diameter of 5 cm was removed; its consistency was gelatinous and its contents were caseous and pearly. Histological examination showed the presence of a granulomatous infiltrate, characterized by giant cells and fine calcifications (Fig. 4), typical of calcifying pseudo-tumor [1, 2, 3, 4, 5]. The postoperative period was uneventful. At 36-month follow-up examination, the patient's clinical symptoms had entirely regressed and there was no evidence of recurrence on either MRI or CT scan.

Discussion

Calcifying pseudo-tumor is a benign, non-neoplastic lesion with a probable inflammatory reactive origin [1]: involvement of the neural axis by this lesion has only recently been described. As we can see in Table 1, which summarizes the 13 cases reported in the literature and the case reported here, the spinal localization can be considered rare, and may represent a real surprise for the neurosurgeon. An analysis of these 13 cases shows that males

Fig. 1 Spin echo T2-weighted magnetic resonance (MR) image obtained in the sagittal plane shows a low-intensity-signal epidural mass involving the right portion of spinal canal with extension to the homolateral foramen

Fig. 2 Spin echo T1-weighted MR image obtained in the axial plane shows a low-intensity-signal epidural mass involving the right portion of spinal canal with extension to the homolateral foramen

are affected more frequently than females (9/13), that the average age of these patients is 46.2 years and that the cervical segment is more frequently involved (6/13) than the thoracic (3/13) and lumbar (4/13) ones. The duration of clinical history varies from a minimum of 4 weeks to a maximum of several years. Clinical symptoms do not have

Fig. 3 Computed tomographic scan shows a calcified epidural mass in the same location

Fig. 4 Optic microscopic appearance. The lesion is composed of a fibrotic area comprising calcifications (*black arrows*) and a cellular area comprising histiocytes and multinucleate giant cells (*black and white arrows*)

specific characteristics, although local pain was reported in 11 cases. The two cases where pain was not reported – the present case and one of those described by Mayr et al. [3] – were characterized by a brief clinical history and symptoms of spinal cord compression and progressive paraparesis. Surprisingly, in our case, a right-sided lesion caused a bilateral spinal cord dysfunction. We hypothesized that the lesion might have induced a hemodynamic disturbance leading to relative ischemia. Although it has been suggested that this lesion originates from an inflammatory reaction, in our case laboratory tests failed to demonstrate the presence of either local or generalized inflammatory processes. The radiological characteristics observed in our case, namely the presence of calcifications on CT scan and the hypointense appearance on both T1-

Table 1 Details of the 13 cases of calcifying pseudo-tumor of the spine reported in the literature, including the present case (*NR* no recurrence, *R* recurrence)

Authors	Sex/Age	Site	Clinical symptoms	Duration	Surgical removal	Follow-up
Bertoni et al. (1990) [1]	M/23	T10 epidural	Thoracic pain	5 years	Subtotal removal	–
	M/58	C2-C3 epidural	Intermittent pain	15 years	Subtotal removal	NR 9 years and 4 months
			Weakness	3 months		
	M/12	C6 epidural	Stiff neck, neck pain	1 months	Subtotal removal	NR 3 years and 3 months
	M/32	L4-L5 epidural	Low back pain	Several years	Subtotal removal	NR 7 years
	F/33	T9 epidural	Thoracic pain	3 months	Subtotal removal	–
	F/68	L4-L5 epidural	Pain	5 months	Subtotal removal	Degeneration lumbar facet joint 16 months
	F/20	C2 epidural	Occasional finding	–	Subtotal removal	–
	F/56	L4-L5 epidural	Low back pain	–	Subtotal removal	–
Moser et al. (1994) [4]	M/68	C7 left epidural	Radicular pain	4 weeks	Total removal	–
Smith and Berry (1994) [5]	M/48	L2-L3 intradural extramedullary posterior	Crampi sciatalgia	4 years	Subtotal removal	–
Mayr et al. (2000) [3]	M/58	T10-T12 epidural anterior	Paraparesis and pain	1 year	Subtotal removal	NR 4 years
	M/63	C3-C4 left epidural postero-lateral	Paraparesis	3 months	Subtotal removal	NR 5 years
Chang et al. (2000) [2]	M/60	C2 intraosseous	Nuchal pain	4 years	Subtotal removal with osteosynthesis	R 2 years
Liccardo et al. (2003) (present study)	M/40	T8-T9 right epidural lateral	Paraparesis and pain	2 months	Total removal	NR 3 years

and T2-weighted images without gadolinium enhancement on MRI, are concordant with the findings reported by other authors [2, 3, 5], with the exception of Moser et al. [4], who describes a hyperintense image on both T1 and T2 weighting. Myelography [1] documents an epidural lesion, at the level of the intervertebral space, simulating the presence of herniated disk tissue. MRI findings of calcifying pseudo-tumor are quite peculiar, since images of epidural lesion usually show a solid mass with marked contrast enhancement after gadolinium [4]. Neuroradiological differential diagnosis should be made firstly with synovial cysts, neurinomas and psammomatous meningiomas, and secondly with epidural abscesses and old calcified hematomas [3, 4]. Histologically, calcifying pseudo-tumor is composed of a fibrocellular network within which there are calcifications (large granules or linear), chondroid material surrounded by histiocytes and multinucleate giant cells [1, 2, 3, 4, 5]. In particular, the complete absence of cellular elements in the proliferative phase testifies to the non-neoplastic nature of the lesion, while the absence of lymphocytes and Langherhans giant cells rules out both a tubercular localization and a bacterial etiopathogenesis [1]. In the

case we treated, the lesion was confined to the epidural space in a right postero-lateral position: the epidural localization of calcifying pseudo-tumor is undoubtedly the most frequent (Table 1), although some authors failed to specify the position of the lesion on the axial plane. Smith and Berry [5] described the only case with an extramedullary subdural localization, while Chang [2] observed an intraosseous spinal lesion. The surgical approach employed in our case, which permitted total removal of the lesion, was a posterior approach extended laterally, as in the other cases reported in the literature. Total excision of the lesion was described in just one case (Moser et al. [4], case 2), while in the remaining cases removal was only partial. Interestingly, the published data indicate that the latter does not influence immediate clinical outcome and long-term prognosis.

Conclusion

The calcifying pseudo-tumor, although it's a rare lesion, has to be taken into consideration when dealing with a slow-

growing spinal calcified extradural lesion, with MRI features of a well-circumscribed non-contrast-enhancing mass. The correct diagnosis may allow an effective treatment even with a limited surgical excision. In fact, the data reported in the literature indicate that excellent long-term

results are achieved even when only a partial removal was possible in order to reduce the risk of complication in the presence of tight radicular adhesences or of an extensive dural infiltration.

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