

Case Report

Tumoral calcinosis involving the cervical spine

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Abstract

Background: Tumoral calcinosis (TC) is a disease of unknown etiology characterized by the presence of calcified masses in the juxta-articular regions of the extremities. Involvement of the cervical spine is very rare. In this report, the characteristics of TC of the cervical spine, including the clinical presentation, radiographic features, and surgical management are discussed.

Case Description: A 90-year-old healthy female suffering from numbness of the upper extremities for 3 months presented with a 2-week history of progressive weakness of the lower extremities. A neurological examination revealed mild weakness and sensory impairment of the bilateral upper and lower extremities. Computed tomography (CT) scans demonstrated amorphous calcified masses posterior to the spinous process that extended into the interlaminar spaces of C3/4 and C4/5. The masses involved the posterior elements of C3-C4. Interestingly, CT scans performed 4 years earlier showed subtle calcification of a yellow ligament at C3/4 and C4/5. However, neither calcified masses nor bone erosion were observed. On magnetic resonance (MR) imaging, the mass showed hypointensity on T1- and T2-weighted images. The lesion was compressing the spinal cord and was resected surgically. The pathological findings were consistent with those of TC. The natural history of TC is not understood. However, this case suggests that calcified masses may progress within several years and that the bone around the mass may be involved. Postoperatively, residual masses may disappear spontaneously, while new bone is formed in the erosive lamina and facet.

Conclusion: The treatment of choice for TC, if the lesion causes progressive symptoms, is surgical resection.

Key Words: Cervical spine, myelopathy, tumoral calcinosis

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INTRODUCTION

Tumoral calcinosis (TC) is a pathological entity with an unknown cause characterized by the formation of calcified masses in the periarticular soft tissue.^[1,2,5,6] This disorder is categorized into three different clinical

classifications: (i) Lesions developing as a complication of renal dialysis, (ii) lesions with a genetic association (patients with TC usually have heritable abnormalities of vitamin D metabolism), and (iii) lesions occurring sporadically in patients with degenerative or inflamed tissue.^[1] TC occurs predominantly in

the juxta-articular regions of the extremities, and involvement of the cervical spine is very rare, with only seven cases having been reported (including ours).^[1,3,5-7] In this case report, we discuss the characteristics of TC of the cervical spine, including the clinical presentation, radiographical features, and surgical management.

CASE REPORT

A 90-year-old female presented with a 2-week history of progressive spastic quadriparesis visited our clinic. The laboratory data showed no remarkable abnormalities.

CT and MRI findings. A computed tomography (CT) scan of the cervical spine [Figure 1] demonstrated an amorphous calcified mass posterior to the spinous process that extended into the interlaminar spaces of C3/4 and C4/5 with encroachment into the spinal canal. The mass involved the posterior elements of C3-5, and the spinous process and facet had become erosive. Interestingly, a CT scan performed 4 years previously [Figure 2] to evaluate neck pain had demonstrated subtle calcification of a yellow ligament at C3/4 and C4/5; however, neither calcified masses nor bone erosion were observed. On magnetic resonance (MR) imaging performed on the current admission [Figure 3], the mass exhibited hypointensity on both T1- and T2-images. In addition, the lesion was encroaching into the spinal canal and strongly compressing the spinal cord.

Operation and postoperative course

Calcification of the nuchal ligament was marked and, as the ligament was resected, leakage of yellow-whitish fluid was observed. In the interlaminar spaces of C3/4, a large calcified mass was observed. Laminotomy was performed at the C3/4 and C4/5 levels, and a large mass was resected. Pathological examinations [Figure 4] showed amorphous calcified deposits (A: Hematoxylin and eosin stain, and B: Von Kossa stain) with CD68-positive macrophages (C: Immunohistochemical staining), suggesting a foreign body reaction. These findings were consistent with a diagnosis of TC.

The patient's postoperative course was good, and a CT scan showed some of the small calcified masses to be residual [Figure 5a]. CT performed 7 months after the surgery demonstrated the disappearance of the residual small calcified masses with new bone formation in the involved lamina or facet of C4 [Figure 5b].

DISCUSSION

TC is a rare disease of unknown etiology characterized by the presence of large calcified masses occurring predominantly in the juxta-articular regions of the extremities.^[1,2,5,6] This disease, as mentioned above, is

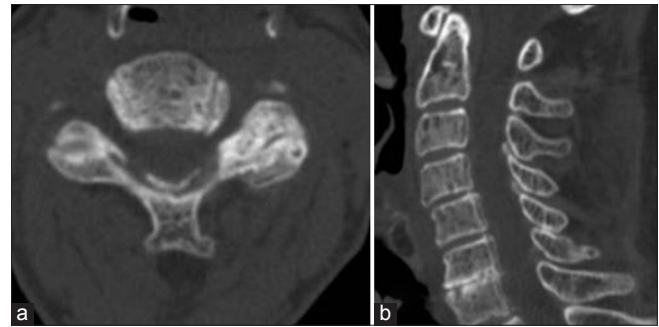


Figure 1: CT scans (a and b) obtained just before the operation showed an amorphous, expansible, multilobulated and calcified mass in the posterior element of C3 and C4, with encroachment into the spinal canal

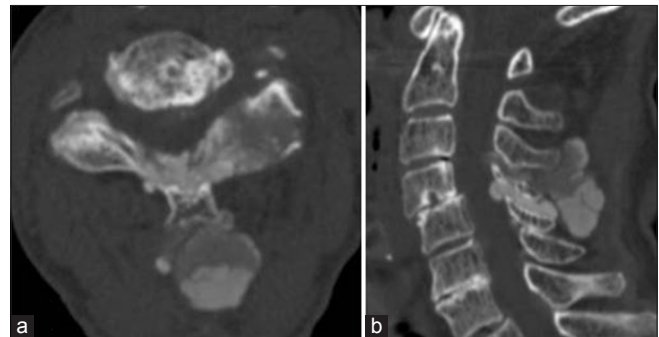


Figure 2: CT scans obtained 4 years prior to the current admission to evaluate neck pain (a and b). Subtle calcification of the yellow ligament was observed at the C3/4 and C4/5 levels

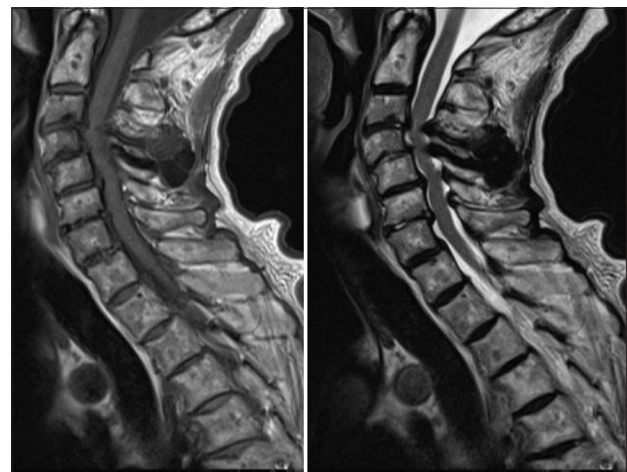


Figure 3: MRI performed prior to the operation showing a large low-intensity mass on both T1- and T2-WI, suggesting the mass to be calcified

divided into three categories. The present case appears to have been idiopathic, and the patient's familial history and laboratory data were unremarkable.

Reported Cases [Table 1].

TC involving the cervical spine is rare, while only seven cases (including ours) having been reported.

The seven reported patients (five females and two males) ranged in age from 1.4 to 90 years.

Two patients presented with occipitalgia, two patients presented with torticollis and three patients developed neurologic signs. Two patients had underlying medical conditions: One reported a history of dialysis and one had Creaste Syndrome. The level affected by the lesion was the upper cervical region in all patients, and the site of the lesion was posterior in six of the seven patients. The locations of the soft tissue around the spine, such as the nuchal ligament, interspinous ligament, and yellow ligament were all posterior.

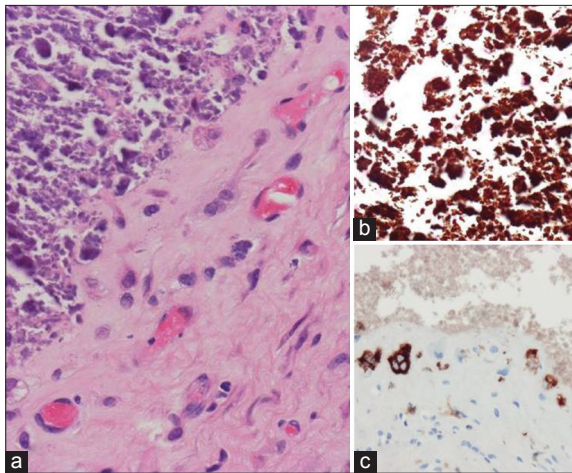


Figure 4: Photomicrographs of the lesion (a) hematoxylin–eosin stain, (b) von Kossa stain and (c) immunohistochemical staining revealing amorphous calcified deposits with CD68-positive macrophages, suggesting a foreign body reaction. These findings were consistent with tumoral calcinosis

Neuroradiology

TC presents with lobulated, calcified, expansible masses on CT scans.^[2,4-7] On MRI, the lesions show a predominately low signal intensity on all sequences. In order to demonstrate the lesion precisely, performing CT is more suitable than MRI. The masses commonly present as growing tumors. However, the progression of a calcified mass involving the cervical spine has been demonstrated in only two cases: Within one year in one case (No. 5)[™] and within 4 years in the current case. TC involves the bone around the mass and may be associated with bony erosion and destruction.^[6] TC may also have a mass effect on neurovascular structures, and bony involvement was identified in five of the seven reported patients. In the present case, residual small masses were observed on CT performed soon after the operation and disappeared 7 months later. Furthermore, new bone formed in the involved lamina and facet.

Methods of treatment

The natural history of idiopathic TC is not well understood. The present case suggests that calcified

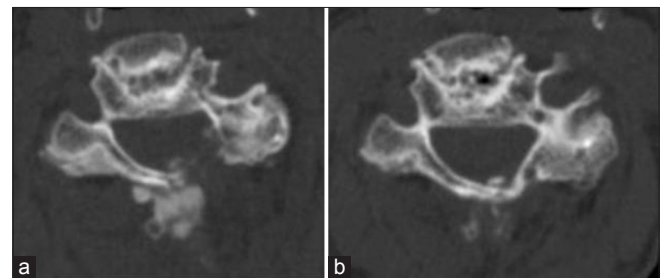


Figure 5: Postoperative CT scans at the C3/4 level. (a) Three days after the operation, residual calcified masses were observed posterior to the lamina of C4. (b) Seven months after the operation, new bone had formed in the involved lamina and facet of C4

Table 1: Summary of cases of tumoral calcinosis involving the cervical spine

Author year	Sex/age	Associated condition	Symptom	Treatment	CT finding			Comment	
					Level	Location	Bone Involve.		Canal Exten.
Kokubun et al. 1996	F/68	np	Nape pain	Removal, laminectomy	C1-C2	Posterior	+	+	A large calcified mass and small masses at C3-7
Ohashi et al. 1996	F/12	np	Torticollis	Excision	Upper to middle cervical	Posterior	+	+	A large lobulated calcified mass no recurrence
Mooney et al. 1997	M/1.4	np	Torticollis	Resection of the mass within the spinal canal	C1-C2	Lateral	-	+	A large partially calcified mass
Teng et al. 2006	F/59	Crete Synd.	Radiculopathy	Resection, laminectomy, arthrodesis	C3-C5	Posterior	+	-	Marked erosive changes of the facets of C3 to C7. progression of bone erosion within a year. An amorphous calcified mass
Miyakoshi et al. 2007	M/54	np	Myelopathy	Resection, laminectomy, arthrodesis	C3-4	Posterior intracanal	-	+	A mass localized in the dura
Tuy et al. 2008	F/50	Hemodialysis	Neck pain	Evacuation	C2-C3	Posterior	+	+	An expansible calcified mass
Ours 2014	F/90	np	Myelopathy	Resection, laminectomy	C3-C5	Posterior	+	+	Amorphous calcified masses CT scans 4 years ago showed a small OYL at C3/4

masses may progress and that the bone around the mass may become involved within several years. The treatment of choice for TC, if the lesions cause progressive symptoms, is surgical removal of the mass.

At the time of an operation we adapted laminotomy, because this procedure seemed to be less invasive for the present elderly patient. The calcified mass extending into the spinal canal through the interlaminar space could be resected and the spinal cord was decompressed.

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