Multilevel bilateral calcified thoracic spinal synovial cysts

Report of 4 cases

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✓Synovial cysts of the thoracic spine are quite rare. Bilateral presentation is even less frequent, and to the authors' knowledge multilevel occurrence and consistent calcification have not been reported so far. The pathogenesis of these cysts is unknown and their histological features have not been studied. They may be overlooked as the cause of myelopathy. The authors report a series of 4 cases of bilateral, multilevel, consistently calcified thoracic synovial cysts. The details of clinical, radiological, and histological findings are presented, along with a review of the literature, and a hypothesis on the pathogenesis of these lesions is formulated based on results of the clinical and pathological studies performed in these patients. *(DOI: 10.3171/SPI/2008/8/5/473)*

Key Words • **histological feature** • **spine** • **synovial cyst** • **thoracic spine** • **thoracic synovial cyst**

Synovial cysts of the thoracic spine are rare, and we found only 20 other reported cases.[2-4,7-9,11] We describe 4 cases involving bilateral multilevel CTSSCs that induced myelopathy. Bilateral presentation is particularly unusual, and we believe that multilevel occurrence with consistent calcification is unique to our cases. Accurate diagnosis is made using MR imaging. Surgery involving laminectomy and removal of the cysts has proven to be a very successful treatment. In this report we aim to heighten awareness of the CTSSC as a possible cause of thoracic myelopathy and to demonstrate the benefits of surgical removal. Furthermore, we seek to provide insight into the pathogenesis of these lesions, for which a definitive explanation has not been ascertained.

Case Reports

The medical records, imaging, and pathological findings in 4 patients (3 women and 1 man) were retrospectively reviewed. These patients presented with myelopathy and were treated surgically over the last 3 years by the senior author (K.I.A.) at the Semmes-Murphey Clinic. The demographic data, clinical findings, involved levels, and outcomes are presented in Table 1. On MR imaging these patients demonstrated compression of the spinal cord by CTSSCs at multiple levels and bilaterally induced spinal stenosis (Fig. 1). A hyperintense signal in the spinal cord was observed on T2-weighted MR images. Patients were treated with laminectomy at the involved levels, and all existing lesions were completely resected in one session. Drilling of the CTSSC with a diamond bur laterally allowed its gentle dissection from the dural sac.

Pathological specimens obtained in all cases showed a synovial lining of the cyst (Fig. 2). Furthermore, a consistent sequence of vascular granulation tissue, fibrous tissue formation, chondroid metaplasia–cartilaginous transformation, and calcification was found (Figs. 3 and 4). All our patients experienced complete symptom resolution after a mean follow-up interval of 8 months, and in each case postoperative MR imaging showed a total decompression of the spinal cord at all levels.

Discussion

Synovial Cysts

Synovial cysts of the spine are intraspinal extradural cysts that arise from facet joints. Synovial cysts have been reported in the lumbar, cervical, and thoracic segments; the

Abbreviations used in this paper: CTSSC = calcified thoracic spinal synovial cyst; MR = magnetic resonance; OLF = ossification of the ligamentum flavum.

TABLE 1

*Characteristics in 4 patients who underwent surgery for CTSSCs**

Case No.	Age (yrs), Sex	Symptoms	Signs	Symptom Duration	Level	Site	Outcome
1	48, M	LE numbness & stiffness, difficulty w/ gait, urinary urgency, sexual dysfunction	sensory level at T-10 w/ decreased sensation for pinprick, light touch, vibration, & partial loss of proprioception in LE LE spasticity w/ 3+ DTRs & bilat clonus LE motor strength 4–/5	6 wks	T-10, T-11	bilat, bilat	complete recovery; normal neurological exam w/in 9 mos
2	54, F	LE weakness & numbness, difficulty w/ gait	sensory level of T-10 w/ decreased sensation to pinprick DTRs 3+ w/ lt-sided clonus LE muscle strength 3+/5	4 wks	T-7, T-9, T-10	midline, bilat, bilat	complete recovery; normal neurological exam w/in 9 mos
3	48, F	midthoracic & low back pain, numbness in feet, shocklike sensation in LE on straightening	no sensory level DTRs 3+ w/ bilat clonus	2 mos	T-9, T-10	bilat, bilat	complete recovery; normal neurological exam w/in 7 mos
4	53, F	LE spasticity, history of myelopathy from cervical spondylosis that improved in UE & worsened in LE 1 yr after cervical decompression & stabilization	no sensory level DTRs 3+ w/ bilat clonus	2 mos	T-2, T-3	bilat, bilat	complete recovery; normal neurological exam w/in 7 mos

* DTR = deep tendon reflex; LE = lower extremity; UE = upper extremity.

majority occur in the lumbar segment, with fewer in the cervical level, and we could only find 20 other reported cases in the thoracic spinal segment.[1–3,9,17,22] The cysts may occur unilaterally or in rare cases bilaterally.[9,17] We believe our cases constitute the first report of multilevel occurrence. The cyst may calcify, although this is rare, particularly for lumbar cysts. It may be filled with clear mucinous fluid or gas, and the walls may be composed of loose myxoid connective or fibrogenous tissue.[3,9]

Thoracic Synovial Cysts

Paraparesis is the most commonly encountered symptom caused by synovial cysts of the thoracic spine. Other common symptoms include an unsteady gait, middle back pain, lower extremity spasticity, and urinary and sexual dysfunction. Preoperative diagnosis of synovial cysts can be accomplished using MR imaging. The cyst appears as an extradural round mass located adjacent to a facet joint. Cord compression and spinal stenosis are also apparent on MR imaging.

Surgery involving bilateral laminectomy of the affected level and removal of the synovial cyst from the dura mater has been the applied treatment. The outcome following surgical removal of the cysts has been quite rewarding. All but one of the other reported thoracic cases were treated surgically, and only one showed no improvement. The majority

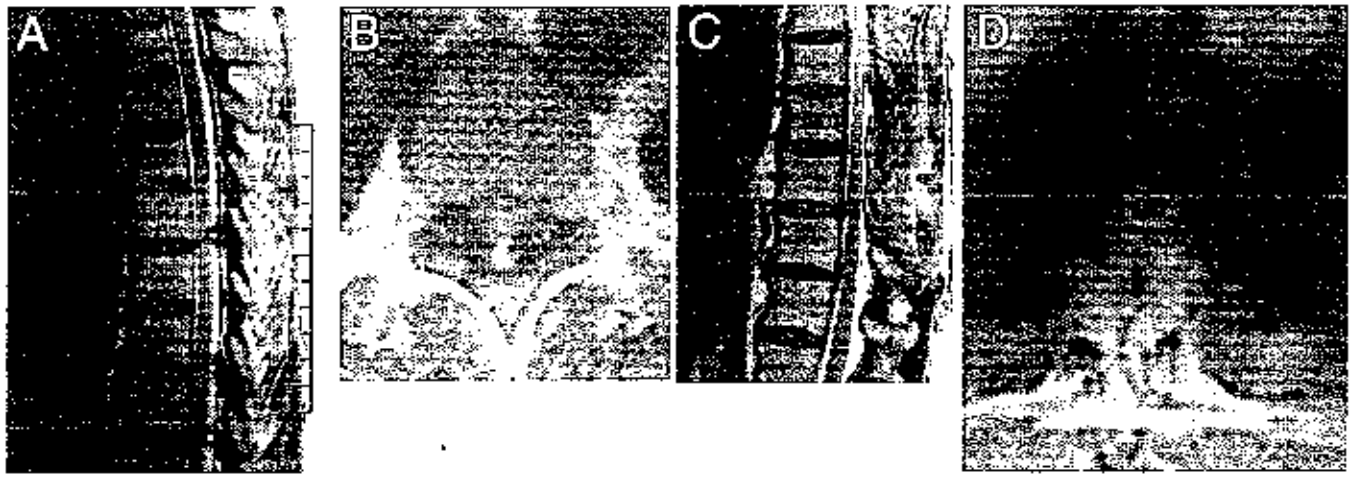

FIG. 1. Case 2. Preoperative sagittal (A) and axial (B) T2-weighted MR images depicting spinal cord hyperintensity (increased signal) at the level of T-7, T-9, and T-10 due to spinal cord compression. Bilateral and multilevel calcified synovial cysts (*asterisks*) are evident. Postoperative sagittal (C) and axial (D) T2-weighted MR images demonstrating resection of cysts.

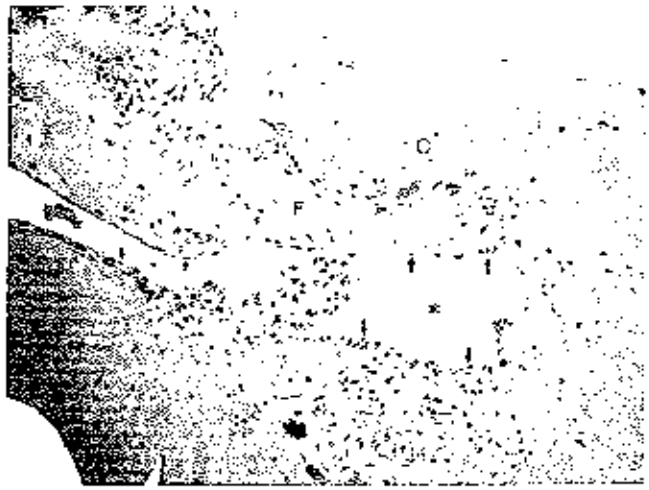

FIG. 2. Photomicrograph showing a cyst (*asterisk*) with synovial lining (*black arrows*), fibrosis (F), focal calcification (*white arrows*), macrophages (M), and few chondroid cells (C). H & E, original magnification × 40.

of patients experience complete symptom resolution or drastic improvement (Table 2).

Differential Diagnosis of CTSSCs

Synovial cysts differ from ganglion cysts in that the former have a true synovial lining (Fig. 2). Ganglion cysts are believed to develop from synovial cysts that disconnect from the adjacent joint. The distinction between synovial and ganglion cysts is not always made in the literature because they have similar clinical and radiological features.[2,7,13]

Thoracic OLF and tumoral calcium pyrophosphate dihydrate deposition disease of the ligamentum flavum produce similar myelopathic features, but have distinguishing anatomical, radiological, and pathological characteristics from CTSSCs. According to our literature review, OLF is mainly reported in the older male population in Japan, with recent reports from China, India, and Africa.[5,9] Dense OLF usually begins laterally and extends to the intralaminar portion of the ligament. Radiologically, dense ossification is depicted on computed tomography scans, with a hypointense signal on T1- and T2-weighted MR images.[5] Pathologically, OLF demonstrates a pattern of endochon-

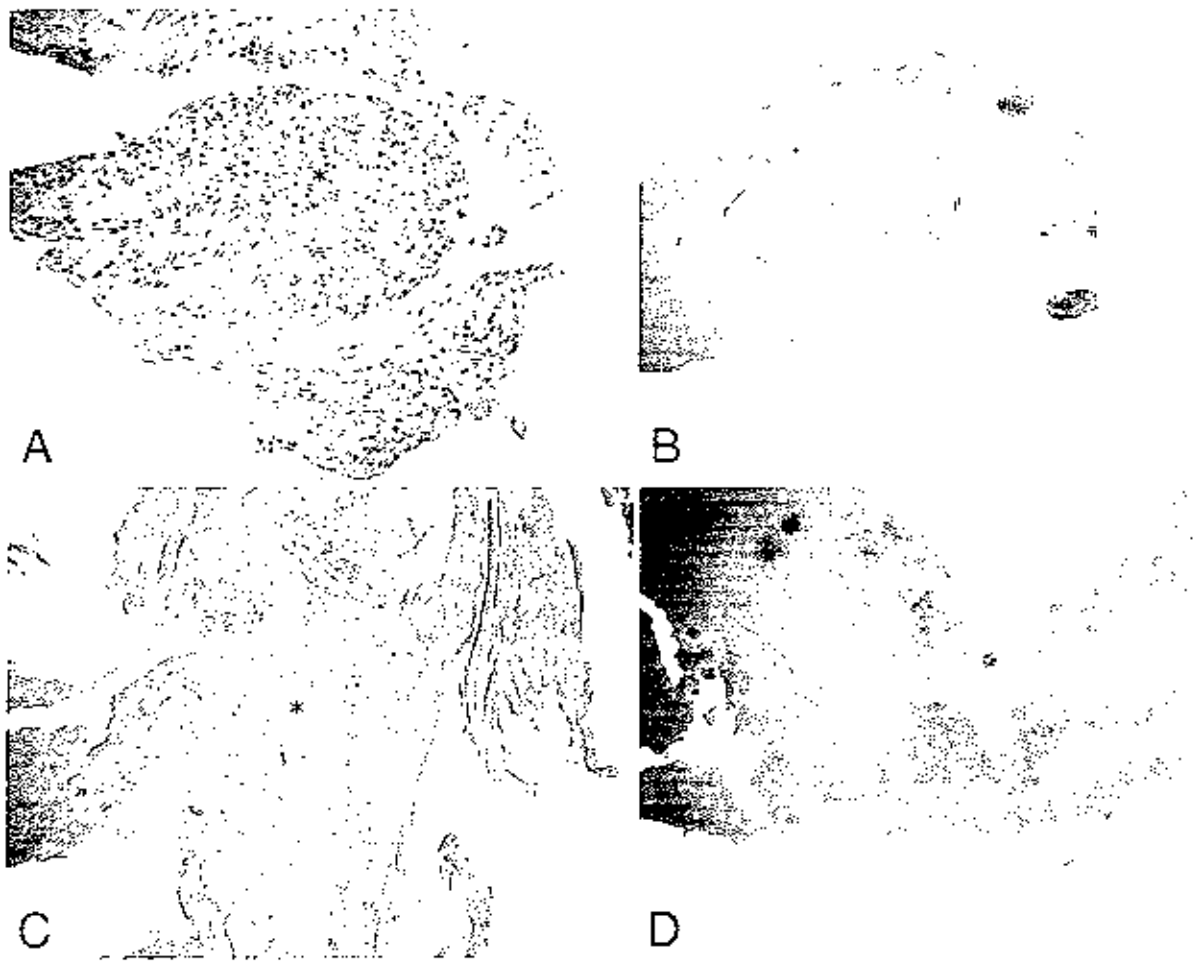

FIG. 3. Photomicrographs demonstrating the proposed stages in the process of pathogenesis in thoracic synovial cysts. A: Vascular granulation tissue (*asterisk*). B: Fibrous tissue (F) and calcification (C). C: Chondroid metaplasia-cartilaginous transformation (*asterisk*). D: Calcification. H & E, original magnification × 100.

FIG. 4. Photomicrograph showing CD34-positive endothelial cells (*dark staining*) in granulation tissue (*asterisk*). Immunostaining, original magnification × 200.

dral calcification of the degenerated ligamentum flavum and well-formed woven bone.[7] Calcium pyrophosphate dihydrate deposition disease of the ligamentum flavum involving the spine tends to occur in the elderly population, is more common among Japanese patients, and shows a female predominance as well as the cervical and lumbar segment preference. Computed tomography scanning shows nodular or ovoid calcified lesions that are continuous with the lamina and are hypointense on both T1- and T2-weighted MR images. They appear in the region of the ligamentum flavum. Histological specimens show crystals with a characteristic rod or rectangular shape, which are embedded in fibrocartilagenous stroma and are birefringent in polarized light.[18]

Hypothesis for Pathogenesis

The most widely accepted hypothesis for the pathogenesis of lumbar synovial cysts involves facet degeneration brought on by various substrates that leads to a weakening in the joint's capsule. This weakening allows a herniation of the synovium, and synovial fluid fills the newly formed cavity and becomes a cyst, which communicates with the associated joint.[5,11] Proposed factors involved in the degenerative process include wear due to excessive movement,[5] spondylolisthesis,[9] and trauma.[1] Alternate proposed mechanisms for synovial cyst development are myxoid degeneration, increased production of hyaluronic acid by fibroblasts in response to repeated stress, and nonspecific

TABLE 2

*Literature review of reported cases of thoracic synovial cysts**

Authors & Year	Patient Age (yrs), Sex	Clinical Findings	Level	Site	Treatment	Outcome
Holtzman et al., 1987	72, M	lumbar pain, radiculopathy; LE weakness & numbness, urinary incontinence	T12–L1, L4–5	bilat	laminectomy	complete resolution
Atwood et al., 1991	59, F	rt sided LE paresis & T10 sensory level	T7–8	unilat	laminectomy	complete resolution
Lopes et al., 1992	35, M	radiculopathy	T10–11	unilat	thoracotomy	complete resolution
Doherty et al., 1993	80, M	LE weakness progressing to complete paraparesis	T9–10	bilat	laminectomy	complete resolution
Fredberg et al., 1993	60, M	LE pain & weakness	T11–12	unilat	laminectomy	complete resolution
	64, F	pain, weakness, sensory loss, & sphincter disturbance	T11–12	unilat	laminectomy	complete resolution
Fritz et al., 1994	50, F	multithoracic & radicular pain, no neurological deficits	T9–10	unilat	laminectomy	complete resolution
Hodges et al., 1994	51, F	thoracic & lumbar pain, no neurological deficits	T4–5	unilat	laminectomy	complete resolution
Hovingaon et al., 1999	NR	radicular pain, spinal cord compression, but no myelopathy	T8–9	unilat	laminectomy	complete resolution
Lynn et al., 2000	24, M	midthoracic pain, LE paresthesias, ataxia	T7–8	unilat	laminectomy	complete resolution
Graham et al., 2001	54, F	LE pain, numbness, & weakness	T11–12	unilat	laminectomy	complete resolution
Cohen-Gadol et al., 2004	73, M	paraparesis, radiculopathy, ataxia, T-10 sensory level	T-10	bilat	laminectomy	improved
	65, M	paraparesis, ataxia, T-1 sensory level, sphincter dysfunction	T-11	unilat	laminectomy	recovered
	68, M	paraparesis, ataxia, T-12 sensory level, sphincter dysfunction	T-11	unilat	laminectomy	improved
	75, M	paraparesis, radiculopathy, ataxia, T-12 sensory level	T-11	bilat	laminectomy	recovered
	81, M	paraparesis, L-1 sensory level	T-11	unilat	laminectomy	recovered
	67, M	paraparesis, T-10 sensory level	T-10	unilat	laminectomy	improved
	76, M	paraparesis, radiculopathy, L-4 sensory level	T-11	bilat	laminectomy	improved
	72, M	paraparesis, LE pain, unsteady gait, L1-2 sensory level	T-12	bilat	laminectomy	improved
	75, M	LE paresthesias, paraparesis, T-12 sensory level	T-12	unilat	laminectomy	no improvement

* NR = not reported.

proliferation of mesenchymal cells.[3] The proposition that synovial cysts occur due to a degenerative process in the lumbar and cervical spine is supported by the fact that the majority of these lesions occur in the most mobile portions and that they tend to occur in an elderly population.

The scarcity of synovial cysts in the thoracic region is believed to be due to the relative immobility of the thoracic spine. Nevertheless, due to this very characteristic of the thoracic spine, we do not believe that increased facet joint motion and microtrauma, as proposed by some authors as the cause of lumbar synovial cysts, is the probable pathogenesis of synovial cysts in the thoracic spine. The majority of the synovial cysts that do occur in the thoracic region appear in the T10–12 interspaces. Because the T10–12 segments of the spine represent the transitional region between the relatively immobile thoracic spine and the highly mobile lumbar segments, it had been postulated that the difference in mobility increases the stress on this segment and accelerates the degenerative changes.[5]

Our cases differ from the majority of other thoracic cases, however, in that we had a slightly younger population, the majority of whom were women; all lesions were bilateral, multilevel, and calcified; and the lesions occurred throughout the thoracic segment. Therefore, the aforementioned hypothesis may not account for all thoracic synovial cysts. Based on the histological findings in the specimens we have obtained, we hypothesize that the inflammatory process originates in the thoracic facet joint and produces vascular granulation tissue (Fig. 3A). This then transforms into fibrous tissue (Fig. 3B), which undergoes chondroid metaplasia–cartilaginous transformation (Fig. 3C), and eventually calcifies (Fig. 3D). The occurrence of neovascularization is supported by the appearance of CD34-positive endothelial cells, which are apparent in the histological specimen (Fig. 4).

Conclusions

Although they are rare, CTSSCs must be considered in the diagnosis of myelopathy. Preoperative diagnosis can be made using MR imaging. The lesions can occur bilaterally and at multiple levels. Surgical removal of these lesions is associated with favorable outcomes and limited recurrence. We suggest that all CTSSCs, bilateral and at multiple levels, could be resected in one surgical sitting. We hypothesize that the pathogenesis of these cysts consists of a capsular inflammatory process, with neovascularization and eventual calcification.

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