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Case Report

Spinal gouty tophus presenting as an epidural mass with acute myelopathy

Che-Kuang Lin, Yi-Hsin Tsai, Shih-Hao Huang, Lin-Hsue Yang, Wen-Jui Chang*

Division of Neurosurgery, Department of Surgery, Far Eastern Memorial Hospital, Banqiao, New Taipei, Taiwan

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ABSTRACT

An uncommon case of spinal gouty tophus was diagnosed in a 74-year-old man who presented in the emergency department with sudden onset of acute paraplegia. The patient underwent laminectomy and nodule removal for neurodecompression. After surgery, the patient demonstrated good functional recovery and returned to baseline performance status. Intraspinal tophi are rare. Image study may show irrelevant findings. In patients with gout, the differential diagnosis should include tophi. In the present case, imaging did not reveal much inflammatory change, but severe symptoms were observed, and a definite preoperative diagnosis was difficult. In cases with neurological compromise, timely neurosurgical decompression leads to good outcomes, as in the present case.

1. Introduction

Gout typically affects the peripheral joints and rarely involves the intraspinal area [1]. It is a common metabolic disorder with well-defined clinical, biochemical, and radiological features [2]. Development of gout involves deposition of monosodium urate monohydrate crystals in the tissues, leading to acute episodes of inflammation and long-term sequelae [2]. Globally, gout affects 1–2% of the population, and the prevalence may increase over time [3]. In Taiwan, the overall prevalence of gout is approximately 3%, with a slight male predominance [4].

Typical images of intraspinal gout include para-articular bony erosions with sclerotic margins. The gouty lesion may show low to intermediate signal intensity on T1-weighted magnetic resonance imaging (MRI) and intermediate to high signal intensity on T2-weighted MRI. The tophus itself is a strong inflammatory agent that causes prominent inflammation affecting the surrounding tissue. Periarticular soft tissue thickening is usual [5].

Patients with spinal gout may present with acute neurological deficits caused by cord or root compression, although this condition is rare. Here, we described a patient with poorly controlled gout that affected multiple peripheral joints; the patient presented with acute myelopathy.

2. Case report

A 74-year-old man presented with a 20-year history of severe gouty arthritis, including tophus formation in the peripheral joints; multiple joint deformities were also noted. In addition, he had suspected gouty nephropathy, resulting in chronic renal insufficiency, for more than 10 years. Two days before admission, the patient experienced an acute gout flare-up, with swelling of the left forearm and fever associated with hyperuricemia (serum uric acid 8.3 mg/dL). During this time, he showed a good response to antibiotic treatment for suspected concomitant cellulitis. However, on the night before admission, the patient developed acute onset of paraplegia with acute urine retention. There was no history of recent trauma, but he reported intermittent lower back pain over the preceding few years. He was unable to walk unassisted for long distances because of severe, bilateral joint deformities in the knees and feet.

Both legs showed negative results in a straight leg-raising test; however, sensory disturbance below the L1 dermatome level was noted. Bilateral deep-tendon reflexes of the Achilles tendon and knee jerk were increased, but the bulbocavernosus and cremasteric reflexes were decreased. His anal tone was sparing but weak. The patient had a distended bladder, and voiding was reported to be difficult; an indwelling urinary catheter was inserted.

Spinal MRI to check for a thoracic or lumbar lesion revealed a 1.2-cm epidural mass at the T12–L1 level, with cord compression

Conflicts of interest: none.

* Corresponding author. Division of Neurosurgery, Department of Surgery, Far Eastern Memorial Hospital, 21, Section 2, Nan-Ya South Road, Banqiao, New Taipei City, Taiwan. Tel.: +886 2 8966 7000; fax: +886 2 2935 6940.
E-mail address: tetsukohayashi@hotmail.com (W.-J. Chang).

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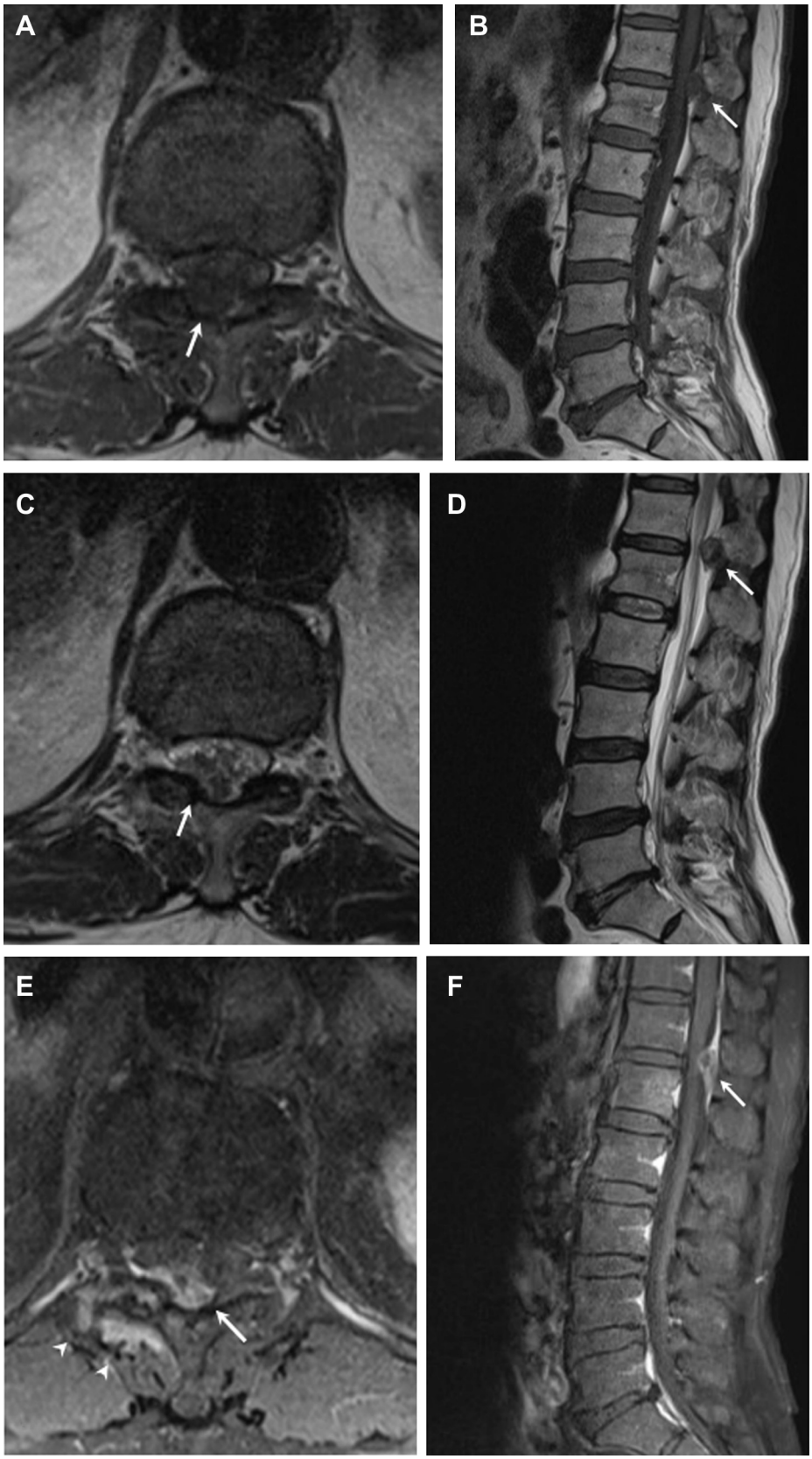

Fig. 1. (A, B) T1-weighted images show an isointense epidural nodule (arrow) with cord compression. (C, D) T2-weighted images show a hypointense epidural nodule. (E, F) T1-weighted images with gadolinium enhancement show rim enhancement of the nodule (arrow) and enhancement of some paraspinal soft tissues (arrowheads).

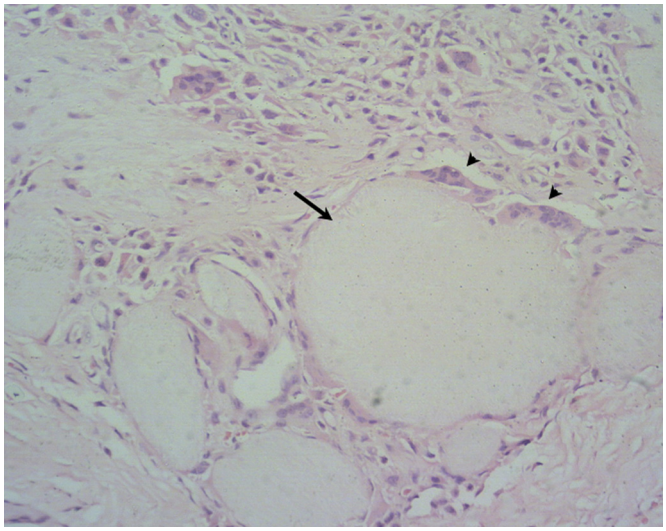

Fig. 2. Foci of tophi (arrow) surrounded by multinucleated giant cells (arrowheads; hematoxylin and eosin stain, 200×).

(Fig. 1A and B). The lesion showed isointensity on T1-weighted imaging and low intensity on T2-weighted imaging (Fig. 1C and D). The cupping appearance of the cerebrospinal fluid surrounding the mass indicated an epidural mass, and postcontrast rim enhancement of the mass with an isointense nodule was noted (Fig. 1E and F). The findings suggested the presence of an abscess or inflammatory changes, but a metastatic tumor could not be ruled out.

Because of the acute myelopathy, emergency laminectomy, and lesion removal were performed. The lesion pathology revealed foci of tophaceous material surrounded by a foreign body reaction and multinucleated giant cells, resulting in the diagnosis of a tophus (Fig. 2).

The patient had an uneventful postoperative recovery, with improvement in paraparesis after surgery. He experienced functional recovery from the paraparesis after a 1-week rehabilitation program and treatment for gout.

3. Discussion

The differential diagnoses of an epidural mass includes synovial cysts, ligament cysts, cystic neurinomas, tumors, hematomas, and abscesses [6]. In our patient, the mass was localized just below the lamina, without involvement of the facet joint or neural foramen; therefore, a nerve sheath tumor was not likely. In addition, there was no postcontrast homogenous enhancement; therefore, a meningioma or a dura-base lesion was unlikely. The peripheral rim enhancement after gadolinium injection was related to an inflammatory process or vascularized granulation tissue [7], although abscesses and tumors may demonstrate similar rim enhancement. An epidural abscess may present as a mass that is hypo- to isointense on T1-weighted images and hyperintense on T2-weighted images, with diffuse homogenous enhancement or rim enhancement. The most common midline extradural tumors of the spine include metastatic tumors, chordomas, medulloblastomas, and ependymomas [6]. A gouty tophus, despite its rarity, should be considered in the differential diagnoses of spinal epidural masses with rim enhancement, especially in patients with systemic gout.

Several reports have described cases of axial gouty arthropathy [5,8–10]. Saketkoo et al [5] described 125 cases of axial gout, including 21 cases of cervical, 19 of thoracic, 50 of lumbar, and 35 of sacroiliac spine involvement, thus demonstrating the predominant involvement of the lumbar spine. The reported symptoms included localized pain, pure radiculopathy, and myelopathy [5]. Many of the

Table 1
Literature reports of cases of spinal gout with myelopathy or cord compression.

Author, year [reference no.]	Age/sex	Location of tophi	Symptoms	Duration	Hyperuricemia	Surgery	Outcome
Koskoff et al, 1953 [11]	44/M	T9–T11	LE paralysis, BBD	6 mo	Yes	T9–T11 laminectomy	Improved
Reynolds et al, 1976 [12]	74/M	L4–L5	Paraparesis	Chronic	Yes	Yes	NR
Magid et al, 1981 [13]	50/F	C7, T2–T9	Back pain, LE weakness, BBD	3 wk	Yes	T1–T9 laminectomy	Improved
Sequeira et al, 1981 [14]	61/M	C3–C4	Quadriparesis, BBD	5 wk	Yes	C3–C4 laminectomy	Improved
Jacobs et al, 1985 [15]	53/M	C1–C2	Quadriparesis	2 mo	Yes	No surgery	Improved
Downey et al, 1987 [16]	73/M	T1	LE weakness, BBD	12 mo	Yes	T1 laminectomy	NR
van de Laar, 1987 [17]	69/M	Cervical spine	LE weakness, myelopathy	1 y	Yes	Yes	Improved
Vervaeck et al, 1991 [18]	56/M	L2–L3	Back pain, paraparesis	3 d	Yes	L2–L3 laminectomy	Improved
Vaccaro et al, 1993 [19]	67/M	C2–C6	Quadriparesis	NR	No	C2–C6 fusion	Improved
Murshid et al, 1994 [20]	79/M	C1–C2	UE pain, quadriparesis	15 mo	No	C1 laminectomy and fusion	Improved
Yasuhara et al, 1994 [21]	60/M	T6–T7	Thoracic myelopathy	3 wk	Yes	T6–T7 laminectomy	Improved
Duprez et al, 1996 [22]	59/M	C3–C6	Quadriplegia	12 mo	Yes	C4–C5 corpectomy	Improved
Pfister et al, 1998 [23]	53/F	T8–T9	Back pain, BBD, LE weakness	3 wk	NR	Yes	Improved
Bret et al, 1999 [24]	59/F	T2–T9	Cord compression	NR	No	T3–T8 laminectomy	Improved
Kaye and Dreyer, 1999 [25]	59/F	T8	Back pain, LE weakness, BBD	18 mo	NR	T8 laminectomy	Stationary
St George et al, 2001 [26]	60/M	T1–T2	LE weakness, BBD	6 wk	NR	T1–T2 laminectomy	Improved
Yen et al, 2002 [27]	68/M	C4–C5	Quadriparesis	2 wk	NR	T9–T11 laminectomy	Improved
Diaz et al, 2003 [28]	74/M	C4–C5	Quadriparesis	1 wk	Yes	C4–C5 laminectomy	Stationary
Dharmadhikari et al, 2006 [29]	66/M	C3–C7	Cord compression, UE and LE weakness, BBD	Several mo	Yes	C3–C6 corpectomy	Died due to multiorgan failure in 1 wk
Popovich et al, 2006 [30]	36/F	T2–T9	Paraplegia	2 wk	Yes	T5–T7 laminectomy	Improved
Lam et al, 2007 [31]	65/M	L3–L4	Acute LE pain and numbness, BBD	4 d	Yes	L3–L4 laminectomy	Improved
Ntsiba et al, 2010 [8]	43/M	T10	Paraplegia	6 mo	Yes	T9–T10 laminectomy	Improved
Levin et al, 2011 [9]	34/M	T1–T5	Paraplegia, BBD	Acute onset	Yes	T2–T10 laminectomy	Improved
Sanmillan Blasco et al, 2012 [10]	71/M	C3–C4	Quadriparesis	4 mo	Yes	C3–C4 discectomy	Improved
Present case	74/M	T12–L1	Paraplegia, BBD	Acute onset	Yes	T12–L1 laminectomy	Improved

BBD = bowel/bladder dysfunction; LE = low extremity; NR = not reported; UE = upper extremity.

reported cases demonstrated positive outcomes after surgical treatment, especially in those with myelopathy (including paraplegia, quadriplegia, and bowel or bladder dysfunction) [8–31] (Table 1).

Typical radiographic and computed tomography findings of chronic tophaceous gout include para-articular bony erosions, often with thin sclerotic margins. The gout may present throughout the structure of the spine and may not be limited to the joint spaces [5]. Gout is a metabolic disorder that results in deposition of monosodium urate crystals and formation of tophi. The tophus itself is a strong inflammatory agent that causes prominent inflammation affecting the surrounding tissue. Most patients with chronic gout have bony erosion at the facet joints in association with periarticular tophi [32]. However, in the present case, an epidural nodule was noted, with rim enhancement but little inflammatory change in the paraspinal muscle and no facet joint involvement on T2-weighted MRI; this finding may serve as a reference for the diagnosis of a mass in the axial skeleton.

Gout is caused by hyperuricemia. Uric acid is a weak acid and is present in the plasma as monosodium urate. At 37 °C, the saturation threshold of monosodium urate is approximately 6.8 mg/dL. Therefore, gout may be cured by long-term reduction in the serum concentration of uric acid (<6 mg/dL) sufficient to dissolve crystalline deposits and prevent the formation of new crystals [2]. Thus, spinal gouty tophi without neurological compromise can be treated medically after complete healing of spinal infections have been confirmed by documented, serial MRI scans [1,15,33]. In addition, many patients with neurological compromise demonstrate good functional recovery if they received proper decompressive laminectomy in time [8–11,13–15,17–24,26,27,30,31] (Table 1).

In conclusion, although axial skeleton involvement is rare in cases of gout, a diagnosis of gout should be considered when a spinal epidural mass is encountered. Medical treatment to lower the serum uric acid concentration may prevent tophus formation. However, timely neurosurgical decompression may benefit patients with epidural gouty tophi and concomitant neurological deficits.

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