



Tophaceous Gout of the Lumbar Spine: Case Report and Review of the Literature

Lomber Omurganın Tofüslü Gutu: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

Andrey VOLKOV¹, David L. RHOINEY², Roderick CLAYBROOKS³

¹Providence Hospital and Medical Center, St. John Health System, Department of Neurosurgery, Southfield, MI, USA

²A.T. Still University-Kirksville College of Osteopathic Medicine, Clinical Affairs, Kirksville, MO, USA

³Providence Hospital and Medical Center, St. John Health System, Southfield, MI, USA

Corresponding Author: David L. RHOINEY / E-mail: drhoiney@atsu.edu

ABSTRACT

Tophaceous gout has classically been described as an affliction of the extremities. It has however been reported as early as 1947 to involve the spinal column. We report a 63-year-old male, previously scheduled for Anterior Cervical Discectomy and Fusion to correct an existing cervical myelopathy at the C3-C4 spinal level, who presented to the emergency room with progressive weakness of the lower extremities and inability to ambulate for three days. Physical examination suggested a possible worsening of his cervical myelopathy but magnetic resonance imaging (MRI) findings remained unchanged from comparison studies. On the day of surgery, he became febrile and complained of excruciating back pain and we therefore initiated an infectious etiology workup and obtained a lumbar spine MRI. Results of imaging suggested a lumbar epidural abscess with effacement of the thecal sac. Emergent L4-L5 decompression led to an evacuation of a "chalky" substance, which was sent for pathology evaluation. This patient was diagnosed with tophaceous gout of the lumbar spine upon final pathological review. We aim to present the management of this case and review the literature associated with this diagnosis with the goal of improving the approach taken to diagnose and treat this pathology.

KEYWORDS: Epidural abscess, Spinal gout, Spinal stenosis, Tophaceous gout, Spinal infection

ÖZ

Tofüslü gut, klasik olarak ekstremitelerin hastalığı olarak tanımlanmakla birlikte 1947 yılında omurganın da tutulumu rapor edilmiştir. Bu olgu sunumunda C3-C4 seviyesinde servikal miyelopati nedeniyle anterior servikal diskektomi ve füzyon planlanan ancak acil servise alt ekstremitelerde progresif güç kaybı ve 3 gündür yürüyememe şikayeti ile başvuran 63 yaşında erkek hastayı sunuyoruz. Fizik muayene hastanın servikal miyelopatisinde muhtemel bir ilerlemeyi işaret ediyordu ancak manyetik rezonans görüntüleme (MRG) bulgularında bir değişiklik olmadığı görüldü. Ameliyat günü hastanın ateşi çıktı ve bel ağrısı ön plana çıktı. Bunun üzerine hastada enfeksiyon kaynağı araştırılmaya başlandı ve lomber spinal MRG yapıldı. Görüntülemenin sonucunda lomber spinal epidural apse ve dural kesede bası tesbit edildi. Acil L4-L5 dekompresyon yapıldı ve tebeşir renkli materyal boşaltılarak patolojik inceleme için gönderildi. Patolojik inceleme sonucunda hastada lomber omurgada tofüslü gut tanısı konuldu. Makalemizde bu olgunun yönetimini sunmayı amaçladık ve ilgili literatürü tanı ve tedavideki yaklaşımı kolaylaştırmak için taradık.

ANAHTAR SÖZCÜKLER: Epidural apse, Spinal gut, Spinal stenoz, Tofüslü gut, Spinal enfeksiyon

INTRODUCTION

Tophaceous gout is a common disease that rarely affects the spinal column. The disease classically has been described as being found exclusively in the extremities, but has also been reported to affect the spinal axis as early as 1947 (1,5). Clinical presentations have been described as mimicking gross pathologies such as epidural cysts, spondylodiscitis, septic arthritis of the facet joints (with or without pseudo-fistulas tracts to the skin) and as epidural abscesses (Table I). However, patients have demonstrated variable clinical signs and symptoms such as myelopathy, radiculopathy, isolated pain, and weakness as reported over the years, making the diagnosis difficult. Diagnosis of tophaceous gout of the spine is based upon histopathological review of an offending specimen and

thus is preceded by treatment of the patient. As reported in the literature, treatment has been primarily in the form of surgical management through a decompressive laminectomy of the affected level with evacuation of the tophus in its entirety and administration of acute anti-gout medication post-operatively. We report a 63-year-old male with a known history of progressive cervical myelopathy and gout, who was scheduled for C3-C4 Anterior Cervical Discectomy and Fusion (ACDF) but became febrile and experienced excruciating low back pain on the day of surgery. Following a thorough workup, the patient was discovered to have findings on magnetic resonance imaging (MRI) suggestive of an L4-L5 epidural abscess with meningeal inflammation and was taken for an emergent L4-L5 laminectomy with evacuation of the offending pathology. Final pathological review of the

Table I: Case Reports Involving Gout of the Lumbar Spine

Author and Year	Age (years)/ Sex	Hx of Gout (Yes/No/N/A)	Vertebral Level	Clinical Presentation	Surgical Management
Current study	63/M	Yes	L4-L5	Progressive B/L LE's weakness	L4-L5 Laminectomy
Lam et al., 2007	65/M	Yes	L3-L4	Right leg weakness w/o LBP	L3-L4 Laminectomy
	63/M	Yes	L4-S1	Bilateral lower limb pain with numbness	L4-L5 Laminectomy, TLIF; L4-L5 pedicle screw fixation
Kelly et al., 2005	56/F	Yes	L4	LBP with radiation to right leg	L4-L5 Laminectomy
Yen et al., 2005	65/F	No	L5-S1	LBP with lower leg numbness and weakness	L5-S1 Decompressive laminectomy
Mahmud et al., 2005	47/M	Yes	L4-L5	LBP with right sided sciatica	L4-L5 Laminotomy
	58/M	No	L4-L5	Sciatica and claudication	L5 Laminectomy
	71/F	No	L4-L5	LBP with bilateral sciatica	L4-L5 Decompression
Hsu et al., 2002	72/M	Yes	L4-S1	LBP w/ radiation to L leg	Decompressive laminectomy
	27/M	Yes	L2-S1	LBP	None
	77/M	No	L3-L5	LBP w/ radiation to R leg	Laminectomy
Barrett et al., 2001	70/M	Yes	L3-S1	LBP plus fever	L5 Laminectomy
Thornton et al., 2000	27/M	No	L3-L4	LBP	Fine aspiration cytology (FNAC)
Mekelburg et al., 2000	60/M	No	L2-L3	LBP	L2,L3 Laminectomy
Paquette et al., 2000	56/M	No	L3	LBP w/ radiation to L leg	Laminectomy
Gines and Bates., 1998	68/M	Yes	L1-L5	LBP plus fever	Laminectomy
King and Nicholas, 1997	37/M	Yes	L5-S1	LBP w/ radiation to L leg	L5-S1 Laminectomy
Bonaldi et al., 1996	76/M	Yes	L3-L4	LBP plus fever & cauda equina syndrome	L3-L5 Laminectomy; L3-L4 discectomy
Miller et al., 1996	80/F	No	L2-L3	LBP w/ radiation to R leg	L2-L4 Laminectomy, facetectomy, foraminotomy
Fenton et al., 1995	60/M	N/A	L4-L5	LBP w/ radiation to R leg	FNAC
	71/F	No	L4-L5	LBP plus claudication	Laminectomy, discectomy
Staub-Schmidt et al., 1995	45/M	Yes	L5	LBP	FNAC
Vervaeck et al., 1991	52/M	Yes	L2-L3	LBP plus cauda equina syndrome	L2-L3 Laminectomy
Arnold, 1998	72/F	Yes	L4	Radiculopathy	N/A
Das Des, 1988	44/M	No	L5-S1	LBP	N/A
Varga, 1985	76/F	No	L5	Radiculopathy	N/A
Lagier, 1983	84/F	Yes	L2	LBP	N/A
Litvak, 1973	73/M	Yes	L3-L5	Lower Leg weakness	N/A
Hall and Selin, 1960	51/M	No	L4-S1	LBP	N/A
Reynold, 1976	74/M	Yes	L4-L5	Paraparesis	N/A

Abbreviations: N/A = unknown, LBP = low back pain, B/L = Bilateral, LE = Lower extremity, FNAC = Fine needle aspiration cytology, Hx= History.

specimen provided a diagnosis of tophaceous gout of the lumbar spine. We aimed to present the management of this case and review the literature associated with the diagnosis with the goal of improving upon the approach taken in diagnoses and treatment of tophaceous gout of the axial spine.

CASE REPORT

A 63-year-old male with a two-week history of progressive cervical myelopathy at the C3-C4 disc level (and previously scheduled for an C3-C4 ACDF) was admitted from the emergency room (ER), following complaints of worsening ataxia, generalized weakness and severe back pain. His medical history was significant for controlled hypertension and gout for which the patient was currently prescribed Lisinopril and Febuxostat (Uloric) respectively. He denied having any past surgeries or use of alcohol, illicit drugs, or tobacco. Musculoskeletal examination revealed 4 out of 5 motor strength in his upper and lower extremities bilaterally with hyperreflexia throughout. The remainder of the physical examination was unremarkable. MRI of the cervical spine revealed no change from previously obtained similar studies but the patient's symptoms continued to progress and he complained of inability to ambulate (Figure 1). The decision was made to proceed with a C3-C4 ACDF in this patient in the hopes of alleviating and improving his clinical symptoms. However, before transfer to the operating room, he complained of severe diffuse arthropathy with back pain in the lumbosacral region and became febrile with a temperature of 39.4° C. Additionally, physical examination of the patient revealed clonus, severe pain with passive range of motion of the lower extremities, and swelling of bilateral lower extremities. MRI of the lumbar spine with gadolinium demonstrated an enhancing epidural mass with severe

effacement of thecal sac at the L4/L5 level with additional dural enhancement noted to be extending inferiorly into the left L4-L5 nerve root sleeve, highly suggestive of an epidural abscess (Figure 2). Surgical intervention was planned and a L4-L5 decompressive laminectomy with foraminotomy was performed. Approximately 10 cc of a white, thick, chalky "cottage cheese" like material was evacuated from the epidural space and sent for pathologic review, which was consistent with tophaceous gout of the lumbar spine. The patient was placed on Vancomycin, Colchicine, and Allopurinol and discharged on the 6th postoperative day with no residual neurologic complaints (Figure 3). (NOTE: the patient underwent a C3-C4 ACDF on post-operative day 3 to address his pre-existing cervical myelopathy.)

DISCUSSION

Gouty tophi development is influenced by several factors including pH level of the surrounding environment, low temperatures, and the presence of a nucleating agent within the synovial fluid to allow crystallization of previously deposited Monosodium Urate (MSU) (3, 8, 9). The classically defined patient with gout has been well described in the literature as a middle-aged male with acute inflammatory monoarticular arthritis of a peripheral joint, most commonly the first metatarsophalangeal joint, elbow, or knee (10). However, crystallization into the facet joints of the axial spine and compression of the thecal sac by a gouty tophus is a rare occurrence (12). Barrett et al. (1) stated prior to 2001 that only 37 cases of gouty arthritis of the spine had been previously reported. According to the literature, spinal tophaceous gout can vary in presentation, ranging from cervical neck pain to acute paraparesis in patients with a previous history of gout (1, 2, 7, 12). Tophi are the chronic manifestations of the clinical disease progression of gouty arthritis. The clinical appearance

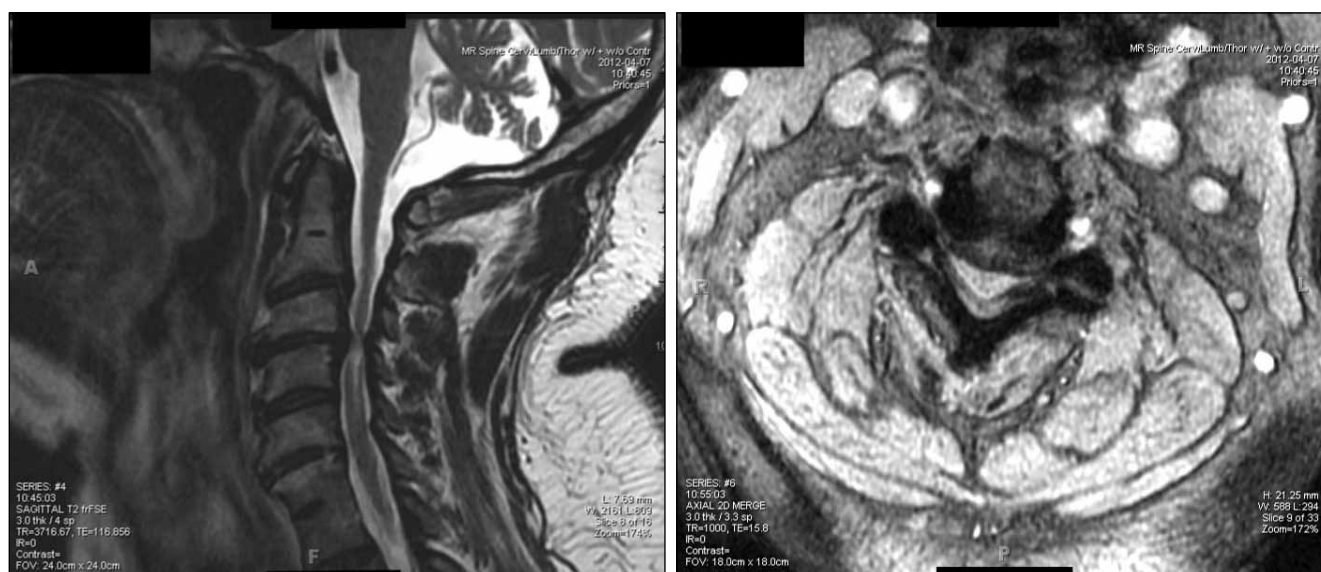


Figure 1: Cervical Spine MRI with and without contrast demonstrating severe cord compression most significant at the C3-C4 spinal level. **Left,** T2 sagittal imaging demonstrating the presence of canal stenosis and cord signal change consistent with a diagnosis of cervical myelopathy. **Right,** axial imaging of the cervical spine demonstrating bilateral foraminal stenosis at the C3-C4 spinal level.

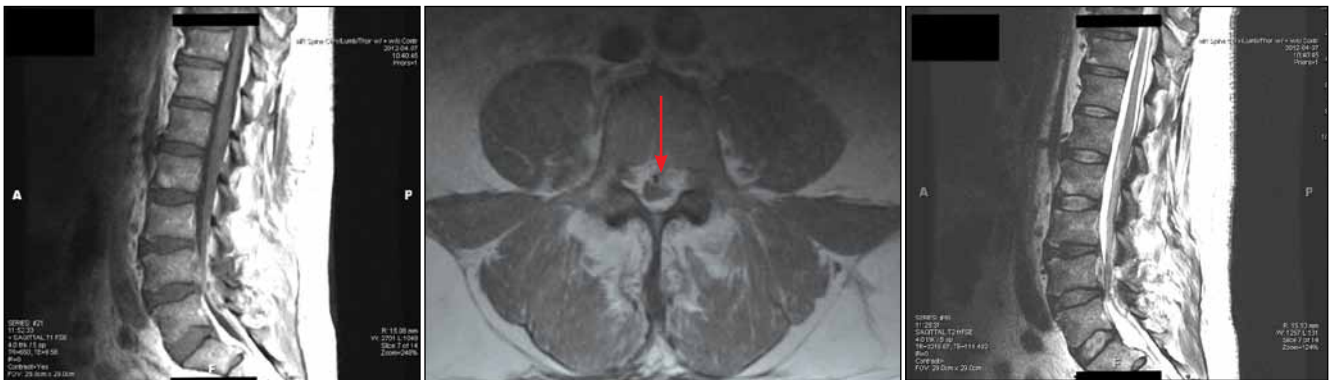


Figure 2: Preoperative MRI with and without contrast imaging of the Lumbar spine. **Left,** T1 sagittal imaging of the lumbar spine demonstrating the offending process at the L4-L5 spinal level. **Middle,** T1 axial images illustrating evidence of a small fluid collection in the anterior epidural space (Red arrow). **Right,** T2 sagittal imaging of the lumbar spine.



Figure 3: Postoperative MRI with and without contrast imaging of the Lumbar spine showing thorough decompression of the thecal sac with some expected postoperative blood products.

and development of tophi in a patient with a known or suspected history of gout typically indicates the presence of chronic gouty arthritis suggesting that the patient has had the disease for at least 10 years (13). It should also be noted that in approximately 30% of untreated gout patients, tophi can develop within five years of onset of the disease (6).

In this patient, formation of gouty tophi in the axial skeleton prior to formation in the larger joints can be explained possibly by poor vascularization within the area leading to the creation of an optimal environment for tophi formation. Given our patient's hyperlipidemic state, obese body habitus and minimal physical activity, the small caliber vasculature in the axial skeleton is hindered, leading to inability to adequately filter the uric acid load deposited in that area. Deposition of uric acid into the interstitial space with decreased filtration mechanisms will eventually lead to

tophi formation. This concept can be extrapolated further by looking at another disease pathology, such as degenerative disc disease (DDD). Lack of nutritional supply to the disc cells is thought to be one of the primary causes of DDD (11). Like all cell types, they require glucose and oxygen to remain active. *In vitro* it has been shown that the activity of disc cells is very sensitive to extracellular pH and oxygen concentration (11). Synthesis rates fall steeply at acidic pH and at low oxygen concentrations. Subsequently, while this type of environment affects the ability of disc cells to synthesize and maintain the disc's extracellular matrix and ultimately leads to DDD, it creates a favorable environment for uric acid deposition and tophi formation. In addition to cervical myelopathy, our patient also suffered from DDD, suggesting that his axial skeleton harbored the environment needed for uric acid formation. This observation may raise a question of whether there is a need for potential screening imagery in patients suffering from gouty arthritis who are in debilitated states or those patients who are also suffering from DDD. This question can only be answered on a larger scale, with randomized prospective studies.

Ultimately, a strong clinical suspicion of the disease is of the utmost importance, as there is no gold standard of diagnosis in the work-up of tophaceous spinal gout. The presumptive diagnosis is made upon clinical signs and symptoms of patient presentation and visualizing MSU crystals on polarized light microscopy eventually confirms the diagnosis. Yu et al. (13) suggested that tophaceous gout could be evaluated by MRI based on signal intensity on T1 and T2-weighted images. This concept was reported by Hsu et al. (4) who showed that periarticular deposits which contain low signal foci on all MRI sequences could possibly be consistent with spinal tophaceous gout. However the differential for this radiological finding also included diskvertebral infection, epidural abscess, rheumatoid arthritis, metastatic disease, dialysis-related amyloid spondyloarthropathy, to name a few. (4) MRI was used to evaluate our patient and as previously stated was most consistent with an epidural abscess at the L4-L5 spinal level. If we ignore our patient's clinical history,

an epidural abscess seems most likely to have caused his acute neurological deterioration. However, patients with a clinical history of gout presenting with acute severe back pain accompanied by radiculopathy or other neurologic symptoms and associated radiological findings suggestive of an epidural abscess warrant a high degree of suspicion for the diagnosis of spinal tophaceous gout. Thus, clinical presentation and history are vital in the workup for this diagnosis. Surgical intervention in the treatment of spinal gout proves to be both diagnostic and therapeutic.

As evidenced in the literature, surgical intervention is primarily in the form of a laminectomy at the offending spinal level (Table I). We performed a similar intervention; however our patient's neurological status remained undeterred postoperatively, likely due to a pre-existing mask of cervical myelopathy or a possible delay in diagnosis (Note: this was later alleviated through a C3-C4 Anterior Cervical Discectomy and Fusion (ACDF)). A disease that has a vast differential diagnosis leaves the surgeon at a disadvantage because often within that group are diagnoses that if left untreated lead to devastating consequences, as well as those which can be treated via less invasive measures. As surgeons treating a patient with an acutely deteriorating neurologic status our natural tendency is to want to "cut." However, in a less critical patient we can be more conservative in our methods. The confirmed tissue diagnosis of spinal tophaceous gout can be obtained via computed tomography (CT) guided needle biopsy or surgical methods. However, we believe that a patient with a known history of gout presenting with infectious like symptoms, severe back pain, and some form of neurologic decline unexplained by a previous diagnosis warrants a thorough imaging studies and exploration, with possible consideration of early surgical intervention.

CONCLUSION

We performed an L4-L5 laminectomy with removal of a "chalky" substance in a patient with a known history of gout who became acutely febrile and then presented with severe back pain and progressively worsening weakness of the bilateral lower extremities. The patient was diagnosed with an epidural abscess based on MRI sequences; however results

of tissue specimens obtained proved to be that of spinal tophaceous gout. We think that the approach presented here warrants further consideration for possible study of the diagnosis and treatment of spinal tophaceous gout.

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