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International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report Spinal gouty tophus presenting as an epidural mass lesion - A case report

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ARTICLE INFO	A B S T R A C T
<i>Keywords:</i> Spinal gouty tophus Epidural mass lesion Hyperuricemia Birefringent crystals Polarized light Case report	Introduction: Gout is a metabolic disease secondary to an increased body pool of urate with hyperuricemia. Gout typically affects the peripheral joints and rarely involves the intra-spinal area. <i>Case presentation:</i> A 43-year-old man, who had metabolic syndrome s/p bariatric surgery and gout suffered from severe left low back pain with radiation to the lateral side of his left thigh and anterior side of his left leg for more than 7 days. His L-spine MRI showed an abnormal posterior epidural space occupying lesion at L4-L5 level. For tissue diagnosis and neural structures decompression, he underwent surgical removal of the epidural mass lesion. The surgical specimen showed a picture of gout and he got a good recovery after operation. <i>Discussion:</i> The differential diagnoses of an epidural mass includes synovial cysts, ligament cyst, cystic neuromas, tumors, hematomas and abscesses. Gout in the spinal canal is difficult to diagnosis, and degenerative spinal diseases. Patients with spinal gout may present with axial pain and a variety of neurological symptoms. <i>Conclusion:</i> Spinal gouty tophus should be considered in the different diagnoses of spinal epidural massee especially in patients with systemic gout. Surgery is needed for final diagnosis. If spinal gouty tophus is highly suspected during the surgery, the specimen should not be preserved with Formalin because birefringent crystals under polarized light is a unique feature for gouty tophus but they dissolve in Formalin.

1. Introduction

Gout is caused either by reduced excretion or increased production of uric acid that results in elevated serum uric acid levels [1]. Hyperuricemia, defined as a serum uric acid level > 7.0 mg/dL, is the most dominant factor in gout development [1]. Gout is an inflammatory disease characterized by the deposition of monosodium urate crystals in joints, cartilage, synovial bursa, tendons or soft tissues with resultant nodules called tophi [1–4]. Crystallization is caused by poor solubility of the synovial fluid compared to the plasma and the lower temperatures exacerbate the phenomenon, so gout usually involves the peripheral joints such as hands, wrists, elbows, feet, ankles and knees [5]. Spinal gout is uncommon; the first report on spinal gout was published in 1950 which was reported by Kersley et al. in a post-mortem examination on a young man [6]. We present a case of spinal gouty tophus presenting as an epidural mass lesion.

This work has been reported in line with the SCARE criteria [7].

2. Case report

A 43-year-old male fisherman, who had metabolic syndrome status post bariatric surgery in 2011, gout since 2000 without regular control, and right side recurrent dacryocystitis came to our Emergency Department in May 2020 with the chief complaint of severe left low back pain with radiation to the lateral side of his left thigh and anterior side of his left leg for more than 7 days. He had visited a local hospital and had taken pain-killers but not effective. On admission, he was afebrile: physical examination demonstrated clear consciousness, multiple tophi over his bilateral elbow joints and bilateral ankle joints (Fig. 1), generalized decreased deep tendon reflex (DTR), antalgic gait due to severe left low back and left lower limb pain but the sensory and motor function of his bilateral lower limbs and his sphincter function were normal. His laboratory data showed elevated level of C-Reactive Protein (CRP) = 18.55 mg/dL and urine acid (UA) = 8.7 mg/dL, otherwise, were within normal limits. Imaging study including plain film and magnetic resonance imaging (MRI) of lumbar spine was arranged. Lumbar spine plain film showed nothing particular but his MRI of lumbar spine

https://doi.org/10.1016/j.ijscr.2021.106063

Received 20 April 2021; Received in revised form 1 June 2021; Accepted 3 June 2021 Available online 11 June 2021 2210-2612/@ 2021 The Authors Published by Elsevier Ltd on behalf of LIS Publishing

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Fig. 1. Joints of this patient. Tophi at the left elbow and bilateral ankles of this patient.



Fig. 2. Pre-operative L-spine MRI.

Upper row: sagittal view, lower row: axial view. A lobulated lesion at the posterior epidural space at the level of L4/5 manifesting non-specific low intensity on T1WI, heterogeneous high intensity on T2WI and with marginal enhancement after gadolinium injection.

disclosed an abnormal posterior epidural space occupying lesion about 2.5cmx0.8cmx0.4 cm in size at the L4-L5 level with the compression of the dural sac and nerve roots. The lobulated lesion manifesting non-specific low T1-weighted image, heterogeneous high T2-weighted image with marginal enhancement after gadolinium injection (Fig. 2).

For tissue diagnosis of the epidural mass lesion and decompression of the dural sac and nerve roots, he underwent total laminectomy of L4 and the complete removal of the epidural mass lesion under general anesthesia. He was put in prone position and a longitudinal incision was made on the midline of his low back after the level of L4/5 was confirmed by taking C-arm film. After the removal of bilateral laminae of L4 and the underlying ligamentum flavum, in the epidural space of L4/5 there was a whitish soft creamy like stuff embedded in the epidural fat tissue (Fig. 3). The epidural lesion was totally excised and sent for both frozen section and Formalin fixed section. The HE stain of Formalin fixed specimen showed the composition of fibrinous exudate, histocytes, fibroblasts and multinucleated giant cells (Fig. 4). The frozen section surgical specimen showed a picture of gout composed of needle shaped crystals which appeared yellow-blue birefringence under polarized lens (Fig. 5). Based on the past medical history, high serum uric acid level of the patient and the specific histological finding of the specimen under polarized lens, spinal gouty tophus was diagnosed. After surgery, his left back pain and the radiation pain resolved and he got a good recovery. During hospitalization, endocrinologist was consulted for the treatment of his hyperuricemia; Colchicine 0.5 mg and Febuxostat 80 mg were prescribed. Probably due to his job, his medication compliance was not



Fig. 3. Intraoperative picture.

The whitish soft creamy like stuff (\rightarrow) was the epidural mass lesion and it was embedded in the epidural adipose tissue(*).



Fig. 4. Histology of the epidural mass lesion, HE stain $600 \times$. The specimen showed the composition of fibrinous exudate, histocytes, fibroblasts and multinucleated giant cells.

good and his follow-up at our out-patient department was irregular; the latest of his serum uric acid level was 7.7 mg/dL checked in November 2020.

3. Discussion

From the Taiwan National Health Insurance Research Database, the estimated prevalence and incidence of gout in Taiwan was 6.24% and 2.74 per 1000 person-years in 2010 respectively, and male patients were affected more [1]. Gouty tophi of the spine are rare. Gout could involve

International Journal of Surgery Case Reports 84 (2021) 106063



Fig. 5. Histology of the epidural mass lesion, frozen section specimen under polarized lens $600 \times$.

Needle shaped crystals which appeared yellow-blue birefringence under polarized lens. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

all anatomic components of the spine including vertebral bodies, pedicles, laminae, ligmentum flavum, interapophyseal cartilage, neuroforamen, epidural and intradural space [8,9]. The most commonly affected level of the spinal gout is the lumbar level followed by the cervical and the thoracic levels [5,9,10]. Patients with spinal gout may present with axial pain, a variety of neurological symptoms such as radiculopathy, myelopathy, cauda equina syndrome, and claudication secondary to cord or root compression [4,8-11]. Spine plain films cannot provide diagnostic value [4,8,10]. Magnetic resonance imaging is still the most important tool of diagnosis; from the previous reports, spinal gout tophi may show isointensity, low or intermediate signal intensity on T1- weighted images and homogenous low intensity to homogenous high intensity or heterogeneous intensity on T2-weighted images. After gadolinium administration, spinal gout tophi may show homogeneous enhancement or heterogeneous peripheral enhancement. The former may reflect vascularized reactive tissue within the tophi, while the latter may result from hypervascular granulation tissue surrounding the tophi [2,4,5,8–12]. Back to our patient, on MRI, his spinal gouty tophus demonstrated as a lobulated lesion at the posterior epidural space at the level of L4/5 manifesting non-specific low intensity on T1WI, heterogeneous high intensity on T2WI with marginal enhancement after gadolinium injection. CT is very helpful to detect the tiny erosions caused by tophi [4,5]. Gout in the spinal canal is difficult to diagnosis before surgery because it is rare and its clinical presentations as well as radiologic findings mimic tumor, abscess, tuberculosis, and degenerative spinal diseases [5].

Hyperuricemia is a major risk factor for gout [1,4]. Patient education pertaining to lifestyle changes and medications are essential to manage hyperuricemia [1]. Medical treatment with colchicine, nonsteroidal anti-inflammatory agents and steroid are effective to control pain in acute attack. Urate-reducing agents are for long-term control, which have two categories. One is xanthine oxidase inhibitors such as allopurinol, febuxostat, and oxypurinol, which are the first line drug of choices for reduced production of uric acid. The other one is uricosuric agents such as probenecid and sulfinpyrazone, which are the second line medications for increasing uric acid excretion [3,5,9,10]. Our patient utilized Colchicine 0.5 mg and Febuxostat 80 mg to treat his hyperuricemia.

4. Conclusion

The differential diagnoses of an epidural mass include synovial cysts, ligament cyst, cystic neuromas, tumors, hematomas and abscesses.

Spinal gouty tophus, despite its rarity, should be considered in the different diagnoses of spinal epidural masses especially in patients with systemic gout. Surgical intervention is needed for final diagnosis. If spinal gouty tophus is highly suspected during the surgery, the specimen should not be preserved with Formalin and should be sent to pathologist in a fresh condition, because birefringent crystals under polarized light is a unique feature for gouty tophus but they dissolve in Formalin.

Sources of funding

No funding was obtained for this study.

Ethical approval

No ethical approval necessary.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors have participated to drafting the manuscript, revised it and approved the final version of the manuscript.

Research registration

Not applicable.

Guarantor

Chi-Man Yip.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

The authors of this article have no conflict or competing interests.

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