CASE REPORT

Severe systemic inflammatory response syndrome immediately after spinal surgery in a patient with axial gout

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SUMMARY

We report a 55-year-old man with gouty arthritis who developed a 3-month history of low back pain, gradual lower extremities weakness and urinary incontinence. Lumbar MRI showed an exophytic lesion at L3–L4. Immediately after spinal decompression surgery, he developed fever, disorientation, polyarthritides, acute kidney injury and leucocytosis. He was treated with multiple antimicrobial agents for presumed spinal abscess but did not improve. Multiple body site cultures were negative. Aspiration of the sacroiliac joint revealed the presence of monosodium uric acid crystals. A diagnosis of acute gout was done, and he was treated with high-dose intravenous methylprednisolone and colchicine. Within 48 hours, he had a remarkable clinical improvement. At discharge, neurological and laboratory abnormalities had resolved. Awareness of risk factors for axial gout and a high degree of suspicion are important to establish a prompt diagnosis and treatment to prevent severe complications as seen in this case.

BACKGROUND

Axial gout was first described in 1949, following the autopsy of a 21-year-old man with partial subluxation of the first cervical vertebrae. At this time, axial gout has been documented in many case reports suggesting that it is not as rare as previously thought.^{1–3} Patients may present with fever, chills, low back pain, radiculopathy, signs of cord compression and even altered mental status.^{1–3} Risk factors associated with axial gout include the presence of peripheral tophi and joint erosions. However, no association has been found with the duration of hyperuricaemia, serum uric acid levels and low back pain.^{4,5} Spinal gout occurs more frequently in the lumbar vertebrae followed by the cervical spine and sacroiliac (SI) joints, with most cases involving more than one vertebral level.⁵ When neurological findings are present, initial treatment consists of surgical decompression along with colchicine, non-steroidal anti-inflammatory drugs or corticosteroids.^{4,6} Recognition of risk factors for axial gout as well as a high degree of clinical suspicion are important to establish a prompt diagnosis and treatment. Herein, we present a 55-year-old man with gouty arthritis who developed a severe systemic inflammatory response syndrome (SIRS) immediately after decompression laminectomy of a lumbar tophus which was initially thought to be an abscess.

CASE PRESENTATION

A 55-year-old man was admitted due to a 3-month history of low back pain, gradual lower extremities weakness and urinary incontinence. A lumbar CT done prior to admission showed changes suggestive of L3–L5 septic arthritis with epidural abscess causing significant canal stenosis. His medical history was pertinent for arterial hypertension treated with losartan 25 mg daily and gouty arthritis treated intermittently with indomethacin and prednisone. He never used uric acid lowering therapy as recommended by his primary physician. He drank two to three vodka shots mixed with high-fructose juice almost daily. He did not use illicit drugs.

INVESTIGATIONS

On admission to our institution, he presented with severe low back pain and marked weakness of lower extremities. Lumbar MRI showed inflammatory changes at the L4–L5 facet joint and an exophytic lesion extending posteriorly from the left L3–L4 facet joint (figure 1A,B). In view of a suspected lumbar abscess, laminectomy and debridement were performed. Vancomycin was started at the time of surgery. The operative report did not indicate any major complications. However, within 24 hours after surgery, his clinical status deteriorated dramatically. The temperature increased from 36.7°C at admission to 38.8°C, heart rate increased from 75 beats per minute to 111 beats per minute and blood pressure decreased from 154/94 mm Hg to 105/70 mm Hg. A marked increase on several laboratory parameters was observed: white cell count from $11.4 \times 10^9/L$ to $18.7 \times 10^9/L$, platelet count from $202 \times 10^9/L$ to $566 \times 10^9/L$, erythrocyte sedimentation rate from 78 mm/hour to 148 mm/hour and creatinine levels from 0.8 mg/dL to 8.4 mg/dL. Also, he developed altered mental status characterised by disorientation in time and place. Piperacillin/tazobactam was added empirically due to his deteriorating clinical state and clinical and radiological findings suggesting an infectious aetiology. Despite multiple antimicrobial therapeutic changes including imipenem, fluconazole and caspofungin, he did not improve. Multiple body site cultures and surgical specimen cultures did not report growth of bacterial or fungal pathogens. Follow-up lumbar MRI revealed persistent contrast enhancement along the posterior aspect of the thecal sac as well as diffuse enhancement of the leptomeninges along the nerves roots.

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Figure 1 (A) Sagittal T1-weighted (W) fat-suppressed image with contrast enhancement at the level of L3–L4. Sequence shows enhancing inflammatory changes within and adjacent to L4–L5 facet joint, including inflammatory osseous enhancement. (B) Axial T1-W fat-suppressed image with contrast enhancement at the level of L3–L4. Sequence is showing exophytic heterogeneously enhancing lesion extending posteriorly from the left facet joint.

One week after surgery, the rheumatology service was consulted due to the development of inflammatory polyarthritis and serum uric acid levels of 16.0 mg/dL. Synovitis was observed in both hands, wrists, elbows, knees and ankles. Subcutaneous nodules were present in the elbows and helix of both auricles. A white thick secretion from the subcutaneous nodule of the elbow was examined by polarised microscopy and revealed negatively birefringent needle-shaped crystals consistent with monosodium uric acid crystals. Radiographs of the right first toe and right knee revealed the appearance of ‘mouse bitten’ erosions characteristic of gout. MRI of the SI joints showed left SI joint widening and surrounding irregular contours (figure 2). The left SI joint was aspirated and

microscopic analysis disclosed uric acid crystals. Furthermore, the operative findings were discussed with the neurosurgeon who described the presumed abscess as a white chalky material. Therefore, the diagnosis of acute polyarticular gout, including axial involvement, was made.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of gout includes other crystal-induced arthritis, such as pseudogout, as well as infectious disorders. Using polarised microscopy, calcium pyrophosphate dihydrate crystals were not observed in samples obtained from aspiration of a tophus and the left SI joint. Furthermore, negative cultures from multiple body sites and the lack of response to broad-spectrum antibiotics did not support the diagnosis of an infectious process. Thus, gout remained as the only feasible diagnosis in our patient.

TREATMENT

He was treated with intravenous methylprednisolone 60 mg daily and colchicine 0.6 mg daily (adjusted for renal function). Within 48 hours, there was a rapid decline in fever, synovitis and resolution of neurological abnormalities. He received 5 days of high-dose methylprednisolone and was continued on colchicine. He was discharged home on colchicine and a tapering regimen of prednisone. Allopurinol was started after 4 weeks. Prednisone was discontinued 2 months later.

OUTCOME AND FOLLOW-UP

The patient was seen at the outpatient clinic 3 months after discharge and was found with serum uric acid levels of 5.6 mg/dL without any recurrence of arthritis or back pain. He stopped drinking alcohol and was compliant with a low purine diet. He was continued on colchicine 0.6 mg daily and allopurinol 300 mg daily.

DISCUSSION

We report a 55-year-old man with history of poorly controlled gout who developed severe low back pain associated with gradual weakness of lower extremities and urinary incontinence. Lumbar CT showed a mass involving the lumbar epidural space that initially was reported as an abscess. Immediately after spinal surgery, the patient developed a SIRS manifested by fever, tachycardia, polyarthritis, altered mental status, acute kidney injury,

Figure 2 Axial T1-weighted fat-suppressed image with contrast enhancement at the level of the sacroiliac joints (SI). Sequence shows abnormal contrast enhancement involving both, sacral and iliac, sides of the left SI joint. There are widening and surrounding irregular contours of the intervening joint space of the left SI joint. Findings are most consistently seen with erosions and with sacroiliitis. The right SI joint has a normal appearance for reference.

leucocytosis and elevated inflammatory markers. He responded remarkably well to systemic corticosteroids.

We propose that during the removal of the spinal tophus, debris of uncoated gout crystals ignited the inflammatory response in our patient. This action triggered the release of microcrystals into the surrounding tissue and circulation. It is known that monosodium urate crystals engulfed by phagocytes cause an intracellular electrolyte disturbance that provokes inflammasome activation with subsequent release of cytokines, notably interleukin-1 β (IL-1 β).⁷ The clinical presentation of our case is similar to that observed in autoinflammatory syndromes mediated by IL-1 β which are characterised by recurrent episodes of fever, disorientation and leucocytosis affecting the joints, skin and serosal surfaces.⁸ Moreover, the release of cytokines stimulates endothelial cells, leading to vasodilatation, increased permeability to plasma proteins and the recruitment of leukocytes into tissues.⁹

There are some case reports describing fever, acute kidney injury, leucocytosis and elevation of inflammatory markers as the initial manifestation of axial gout.^{2 10–12} In most cases, surgical intervention resulted in clinical resolution.^{2 12–14} Conversely, our patient developed a severe inflammatory response syndrome that ensued after surgical intervention.

Other crystal-induced arthropathies involving the spine causing systemic manifestations have been described. For instance, in crowned dens syndrome, calcium pyrophosphate or hydroxyapatite crystals are deposited in the atlantoaxial region of the cervical spine.¹⁵ Aouba *et al* reported eight cases with crowned dens syndrome who presented with fever, headaches, chronic neck pain and a systemic inflammatory response. Some patients were initially diagnosed to have polymyalgia rheumatica, giant cell arteritis or meningitis.¹⁵ The diagnosis of crowned dens syndrome was made by imaging studies and laboratory workup. Similar to our case, all patients responded remarkably well once started on treatment for acute crystal arthropathy. It is known that both, gout and calcium pyrophosphate deposition disease (CPPD) arthritis, elicit their inflammatory response by engaging the recruitment of leucocytes and release of cytokines as discussed before.

Radiographic modalities such as CT and MRI are central in the diagnosis of axial gout. More recently, technical advances in dual-energy CT have further improved the usefulness of radiological images in the diagnosis of gout as it identifies uric acid deposits with a higher degree of specificity than the standard CT or MRI.¹⁶ Studies analysing data collected using newer radiographic modalities reported that 30% or more of patients with chronic gout have vertebral uric acid deposits, most of them asymptomatic.^{4 5}

Nonetheless, the diagnosis of axial gout is still challenging. A clinical presentation of fever, hypotension, leucocytosis and elevated inflammatory markers mimic infectious diseases. Similar to our case, there are reports of axial gout being treated initially as an abscess.¹³ High suspicion has to be kept in mind by the surgical team when sending samples for pathological analysis. Routinely, specimens are placed in a formalin medium causing the dissolution of uric acid crystals. Instead, alcohol should be used as a fixative for preservation of gout crystals.¹⁷ In our case, the pathology report did not disclose uric acid crystals as the surgical tissue was fixed in formalin. The diagnosis of axial gout in our patient was confirmed by the presence of uric acid crystals obtained by aspiration of a tophus and the left SI joint.

Guidelines for the treatment of acute gout with axial involvement are not well established. The American College of Rheumatology guidelines for acute peripheral gout recommends

colchicine, systemic glucocorticoid or non-steroidal anti-inflammatory drugs as monotherapy or combination treatment depending on the severity of symptoms.¹⁴ It has been proposed by Hou *et al* that surgical intervention in axial gout should be reserved for those cases with progressive neurological deficit or patients who have failed pharmacological treatment.¹⁸ At the moment, there are no recommendations for presurgical medical management of axial gout. Prophylactic gout treatment with colchicine before surgery could decrease the risk for systemic inflammatory response due to the release of uric acid crystals, and consequently inflammatory cytokines.

In summary, the development of SIRS after surgical excision of a spinal tophus in patients with axial gout must be recognised as a potential complication. Our case describes another perspective of axial gout with an inflammatory response after surgical procedure. Early recognition of axial gout and medical treatment prior to surgical intervention may reduce postsurgical complications.

Learning points

- Patients presenting with a history of tophaceous gout who develop lower back pain should be evaluated for axial gout.
- Surgical removal of a spinal tophus may induce an acute inflammatory response syndrome.
- Early recognition of axial gout and medical treatment prior to surgical intervention can reduce postsurgical complications.

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