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Spinal Abscess Caused by *Salmonella* Bacteremia in a Patient with Primary Myelofibrosis

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
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Conflict of interest: None declared

Patient: Male, 50
Final Diagnosis: Spinal cord compression associated with spinal abscess
Symptoms: Diarrhea • fever • low back pain
Medication: —
Clinical Procedure: Laminectomy
Specialty: Hematology

Objective: Rare co-existence of disease or pathology
Background: In Primary Myelofibrosis (PMF; a clonal disorder arising from the neoplastic transformation of early hematopoietic stem cells) patients, spinal cord compression (SCC) is a common complication or even a presentation symptom due to extramedullary hematopoiesis (EMH). However, a case of SCC caused by a spinal abscess is unusual. To the best of our knowledge, this is the first case report of this rare condition.
Case Report: We are reporting the case of a 50-year-old male with primary myelofibrosis and long-standing splenomegaly with back pain as a presenting symptom who was found to have spinal cord compression. An MRI was performed, as EMH was suspected. The blood cultures revealed an infection with *Salmonella*, so the patient was placed on ceftriaxone, with no response. The patient demonstrated substantial clinical improvement after 2 weeks of neurosurgical intervention and pain management.
Conclusions: In PMF patients, back pain with fever or mild neurological symptoms needs to be investigated urgently because of the high risk of irreversible spinal cord damage leading to partial or complete loss of functional independence and shortened survival. The compression could be related to EMH or infections due to an immunodeficiency.


MeSH Keywords: Epidural Abscess • Primary Myelofibrosis • Salmonella Infections • Spinal Cord Compression

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/903482>

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Background

Salmonellae are gram-negative bacilli of the Enterobacteriaceae family (2 species of *Salmonella*, *Salmonella bongori* and *Salmonella enteric*, are further divided into 6 subspecies that include over 2500 serovars), known to cause a variety of common illnesses from gastroenteritis, typhoid fever, and bacteremia to the asymptomatic carrier state [1,2]. An uncommon complication of *Salmonella* infection is spinal abscess in combination with PMF. According to 2015 Infectious Diseases Society of America (IDSA) clinical practice guidelines for the diagnosis and treatment of native vertebral osteomyelitis in adults, ciprofloxacin PO 500 mg q12 h or IV 400 mg q12 h is the first choice and ceftriaxone 2 g IV q24 h (if nalidixic acid resistant) is the alternative, both for 6–8 week duration [3].

Primary myelofibrosis (PMF) is a myeloproliferative neoplasms (MPNs) with an international incidence of PMF of approximately 1 per 100 000 per year [2].

We are reporting a 50-year-old male with primary myelofibrosis and long-standing splenomegaly associated with back pain as a presenting symptom, who was found to have spinal cord compression.

The mechanism of extramedullary hematopoiesis is believed to be linked to the constitutive mobilization of CD34+ cells into the peripheral blood. This dysregulation of hematopoietic stem cell (HSC) trafficking likely ultimately leads to the seeding of extramedullary sites [1]. Injection of CD34+ hematopoietic stem cells has been clinically applied to treat various diseases, including spinal cord injury [4].

The case was challenging since spinal cord compression in the setting of PMF usually occurs due to extramedullary hematopoiesis. However, in this case, the patient had *Salmonella*

bacteremia and spinal cord compression due to an abscess formation.

Case Report

Our patient was a 50-year-old male with no chronic medical illnesses who presented with fever, diarrhea, and severe lower back pain for 1 week and was unable to move his left leg, which he attributed to severe pain. He had no history of trauma or weight loss.

The physical exam revealed a lower limb power of 5/5, increased bilateral tone, brisk reflexes, bilateral downgoing plantars with no sensory deficit, positive results for the straight leg raising test for the left lower limb, and a tender L5-S1 vertebra. These results raise the suspicion of possible spinal cord compression.

Blood culture revealed infection with *Salmonella* in the blood of the patient. Identification showed *Salmonella* type D. He was started on ceftriaxone 2 g IV q24 h for 6–8 weeks, based on the culture and sensitivity. A spine MRI was conducted and revealed a disc prolapse, and the neurosurgeon advised conservative treatment. The complete blood count showed WBC 9×10^3 HB 10 gm/dl platelets 173×10^3 , a peripheral smear presented a leukoerythroblastic picture, and an ultrasound revealed the presence of massive splenomegaly (20 cm). A positive JAK2 V617F mutation was also noted.

His bone marrow examination confirmed the diagnosis of primary myelofibrosis (Figures 1, 2). The patient fulfilled the 2016 WHO criteria for PMF. We planned to start him on JAK2 inhibitors for the symptomatic splenomegaly.

Due to unresolved pain and fever, the MRI was repeated and showed an epidural mass at the level of the L4–L5 disc. From

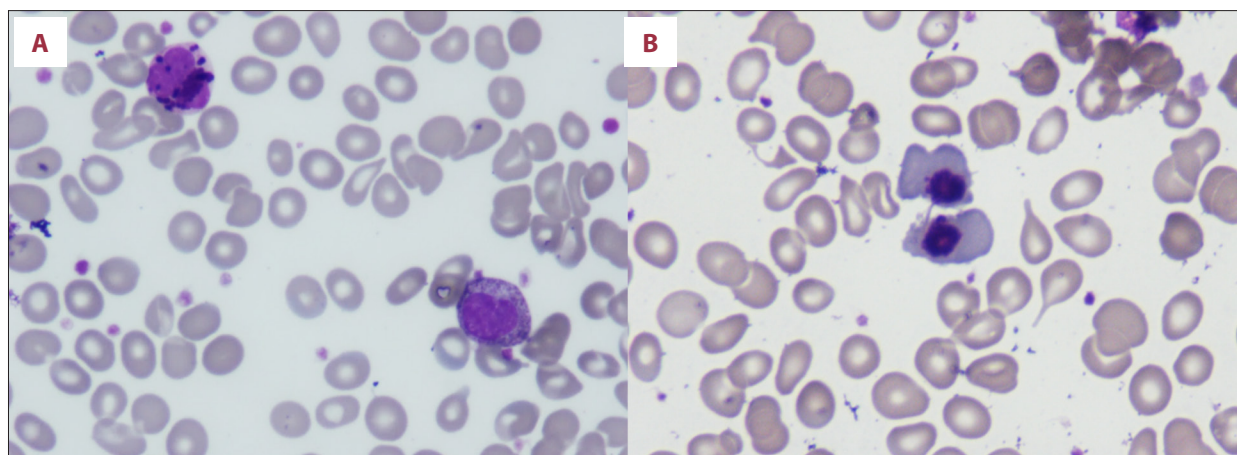


Figure 1. (A, B) Peripheral smear showing neutrophilic leukocytosis with a shift to left and basophophilia (lower left corner) tear-drop cells and leukoerythroblastic picture ($\times 500$).

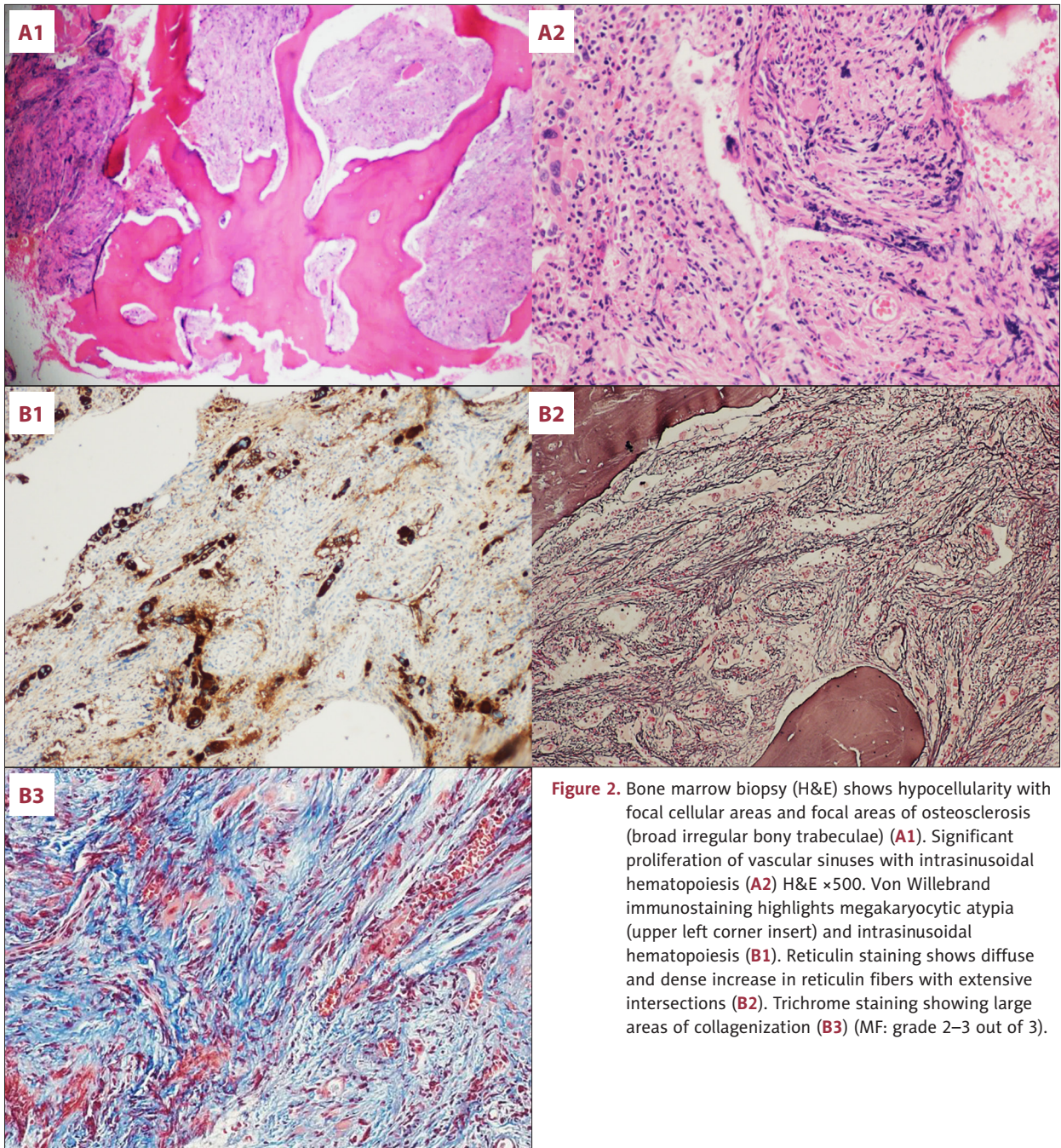


Figure 2. Bone marrow biopsy (H&E) shows hypocellularity with focal cellular areas and focal areas of osteosclerosis (broad irregular bony trabeculae) (A1). Significant proliferation of vascular sinuses with intrasinusoidal hematopoiesis (A2) H&E $\times 500$. Von Willebrand immunostaining highlights megakaryocytic atypia (upper left corner insert) and intrasinusoidal hematopoiesis (B1). Reticulin staining shows diffuse and dense increase in reticulin fibers with extensive intersections (B2). Trichrome staining showing large areas of collagenization (B3) (MF: grade 2–3 out of 3).

the MRI, we suspected an abscess or extramedullary hematopoiesis. The case was discussed with the infectious disease team, as well as the neurosurgeon, who agreed to treat the patient with antibiotics for 2 weeks, and if there was no improvement, to repeat the MRI. The MRI was repeated after 2 weeks and showed evidence of spondylodiscitis at the L4-5 level, with multiple paraspinous abscesses that had increased in size (Figure 3). The neurosurgery team intervened this time because clear pus was aspirated from the epidural mass, and a laminectomy was performed. The patient recovered well after

the procedure, with complete relief of his pain symptoms, and a further MRI showed dramatic improvement (Figure 4).

Accordingly, we decided to regard the organism as *Salmonella* group D, as there is significant cross-reactivity while doing serotyping. For example, *S. enteritidis* (the most likely cause in our patient, based on epidemiological distribution) and *S. typhi* are both under group D and *S. typhimurium* and *S. paratyphi* B are under group B. Further genome sequencing is required to confirm the organism.



Figure 3. Lower, thoracic, and lumbar spondylodegenerative changes. L5–S1 posterior disc protrusion. Posterior disc protrusion at L4–5 level. Reduced height of L4–L5 disk is seen, denoting partial destruction by the inflammatory process. Mass effect on the thecal sac, which appreciably diminished in Figure (B). The lumbar lordosis is straightened in Figure (B) compare to Figure (A) with mild grade 1 spondylolisthesis at L4–L4 level.

In our labs, after positivity of blood culture in the BACTEC blood culture system sample was taken to MALDI-TOF, where identification was done, drug susceptibility was assessed through Phoenix susceptibility testing, and then the sample was serotyped to identify the *Salmonella* groups.

Ceftriaxone 2 g IV q24 h was selected after the result of identification suggested increasing resistance to fluoroquinolone, and after the susceptibility result, we continued with ceftriaxone due to the excellent tissue concentration for such complicated bacteremia and once-daily dosing.

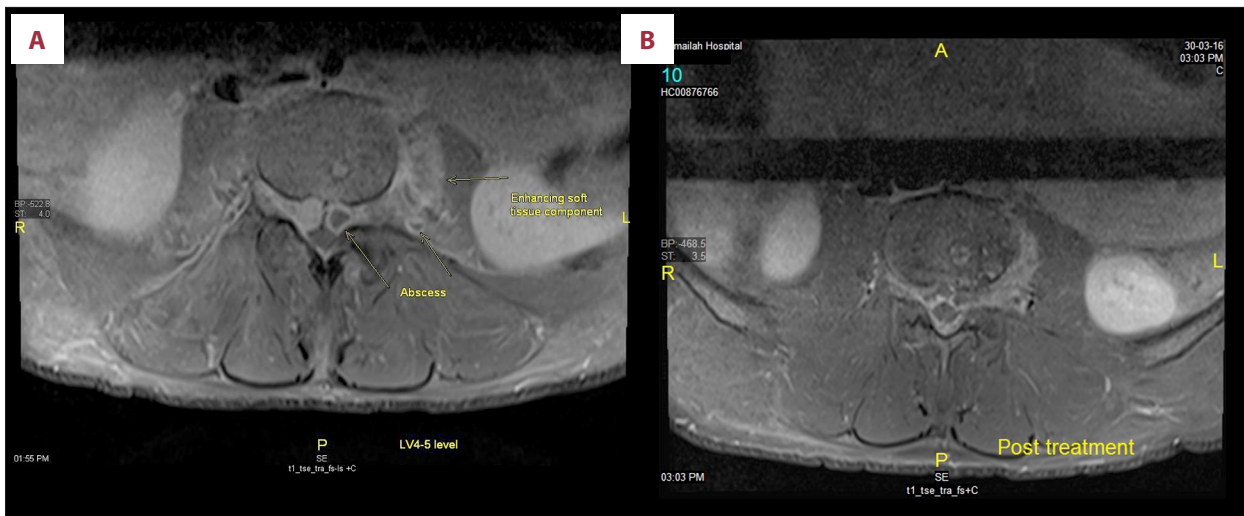


Figure 4. The anterior epidural collection (abscess) with marginal enhancement in Figure (A) measuring 7.9 mm thickness. Same collection measured only 2.1 mm in Figure (B). Decreased left paraspinal edema and fluid collection are noted in Figure (B).

Based on the above evidence and due to the macroscopic description of the abscess, microscopy showing profuse polymorphonuclear cells, and the fact that no other organism was detected in all sepsis work-up performed in our patient, we speculated that the epidural abscess was related to *Salmonella* bacteremia. Epidural abscess culture failed to grow any organism, probably because our patient was on an antibiotic for more than 2 weeks before drainage.

Discussion

Spinal cord compression is a hematological emergency, leading to morbidity and mortality if not treated promptly with appropriate and accurate measures. It can occur due to infiltration or compression by a mass. The compression itself can happen due to extramedullary hematopoiesis [5] or infections [6].

Myeloproliferative neoplasms (MPNs) are clonal hematopoietic stem disorders characterized by a high rate of actual proliferation of 1 or more cell lineages. MPNs are overlapping syndromes that can progress to the fibrotic stage or acute leukemia. One of the recognized complications is EMH, which can lead to cord compression.

Our patient presented with back pain but no paraparesis, one of the most common presenting symptoms reported in the literature [6]. The first images from the MRI epidural showed the epidural mass, which could be EMH or infectious.

The patient had MF, which renders EMH with spinal cord compression as a presenting feature a possibility [7]; however, in the presence of *Salmonella* D bacteremia, other options should be considered (e.g., an abscess).

Nevertheless, a large study of more than 500 patients with MF and massive splenomegaly concluded that severe infections were frequent and potentially fatal events [8].

Extraintestinal infections (e.g., endocarditis, arteritis, meningitis, ventriculitis, pneumonia, arthritis, pyomyositis, and cholecystitis, as well as splenic, liver, and urogenital abscesses) have been documented in the literature [9].

Bone involvement accounts for less than 1% of cases. Risk factors such as sickle cell anemia (SCA), being male, connective tissue disease, and immunosuppression have been described. The femur, tibia, humerus, and lumbar vertebrae were the most common sites and usually required prolonged antibiotic courses of more than 4 weeks combined with surgical intervention to produce good treatment results [10]. Our patient required a total of 10 weeks of intravenous and then oral antibiotics combined with a right L4 laminectomy and abscess drainage.

Spinal cord abscesses due to infections are not uncommon. A large meta-analysis using R with around 1000 patients showed impressive results. The youngest patient to be reported was less than 10 years old, and the male-to-female ratio was 1: 0.5. Diabetes mellitus (DM) and a current infection were the most common risk factors. The most common site of the epidural abscess was the thoracic vertebrae, followed by the lumbar vertebrae [8]. Fever was observed in 61% of the cases and back pain in 71%, while paraplegia was seen in 30% of cases.

In a large meta-analysis of 915 patients with spinal epidural abscess conducted by Reihnsaus et al. [6], we only found 3 cases of epidural abscesses secondary to *Salmonella* species like we found in our patient. None of the patients had the

Table 1. List of published cases of spinal cord abscess (listed chronologically).

Case	Sex/age	Organism	Sensitivity	Location	Risk factor	Outcome
Patrick 2003 [11]	6 M	<i>S. enteritidis</i>	Sensitive	Frontal	Sickle cell anemia	Good condition, needed plastic surgery
Abdullah 2004 [12]	56 M	<i>S. eneterica</i>	Sensitive	Cervico-thoracic	DM	Good condition
de Araju 2012 [13]	69 F	<i>S. enteritidis</i>	NA	Thoracic	SLE	Complete recovery
Khoo 2016 [14]	57 transgender	<i>S. typhi</i>	Sensitive	Thoracic	Trauma	Good condition
Present case	50 M	<i>S. typhi</i>	Sensitive	Lumbar	MF	Good condition

DM – diabetes mellitus; SLE – systemic lupus erythematosus; MF – myelofibrosis; NA – not available.

myeloproliferative disorder. The Table 1 shows all cases reported in the literature, including this one.

Conclusions

Spinal abscess is a serious condition that can lead to spinal cord compression and it can be a presenting symptom. There

are several risk factors identified in the literature. Myelofibrosis is another hematological condition that can also present with spinal cord compression. Our patient had both, which made the case more challenging, considering the importance of differential diagnosis and early interventions.

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