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Case Report

A rare case of *Streptobacillus moniliformis* epidural abscess requiring neurosurgical decompression

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ABSTRACT

Background: *Streptobacillus moniliformis* is the primary causative agent of rat bite fever, an infectious disease transmitted through contact with rats through bites, scratches, or exposure to excrement. Before this report, only two instances of spinal epidural abscess (SEA) due to *S. moniliformis* infection have been documented. We present the case of a 76-year-old male who developed a cervical SEA secondary to *S. moniliformis* infection, requiring neurosurgical decompression of the spinal cord.

Case Description: A 76-year-old male presented to the emergency department with bilateral shoulder and back pain, upper extremity weakness, left hip pain, and left thumb pain. He denied any recent exposure to pets or animals, and the initial workup did not yield the source of the infection. Enhanced magnetic resonance imaging of the cervical spine demonstrated C6–7 discitis/osteomyelitis and an associated ventral SEA, as well as discitis/osteomyelitis of the C2 vertebral body and C5–6 endplates. Subsequently, the patient underwent a C3–7 laminectomy and received a 6-week postoperative course of intravenous ceftriaxone, resulting in complete resolution of the abscess. Blood tests revealed the presence of *S. moniliformis*, which the patient attributed to potential rat exposure at his workplace.

Conclusion: Identification and diagnosis of *S. moniliformis* infection requires a high index of suspicion. Neurosurgeons should consider this rare pathogen in the differential diagnosis of SEA to facilitate early detection, diagnosis, and surgical intervention, ultimately improving patient outcomes.

Keywords: Cervical spinal epidural abscess, Rat bite fever, Spinal decompression, *Streptobacillus moniliformis*

INTRODUCTION

Rat bite fever (RBF), caused by the *Streptobacillus moniliformis* bacterium, is an uncommon infectious disease pathogen commonly transmitted through exposure to and accidental consumption of rodent droppings. Less common still are cases of *S. moniliformis* spinal epidural abscesses (SEA); only two cases have been previously reported, one in the thoracolumbar spine and another in the lumbar spine. Herein, we describe a cervical RBF SEA requiring neurosurgical intervention, the first reported in the literature to our knowledge.

CASE REPORT

A 76-year-old male with no significant medical history presented through the emergency department (ED) with new-onset bilateral shoulder and back pain. The patient described himself

as a healthy individual at baseline. He was an active runner and had not had reason to see a physician in years. He reported that the pain started in his right shoulder about 1 week before admission and spread to his upper back and left shoulder. These pains continued to progress throughout the week despite over-the-counter non-steroidal anti-inflammatories. Eventually, he began to develop indolent weakness; by the time of evaluation, he was unable to lift any objects over his head. He then noticed left hip pain that made ambulation difficult as well as new left thumb pain, prompting him to present through the ED. He denied sick contacts, recent travel, outdoor expeditions, recent unclean or public water exposures, and pet or animal exposures. He was subsequently admitted to the medicine service for a diagnostic workup.

The patient was afebrile and hemodynamically stable on room air. He had a cryptogenic leukocytosis with elevated inflammatory markers (C-reactive protein of 353 and erythrocyte sedimentation rate of 87) and a concomitant acute kidney injury. A urinalysis, chest, hip, and shoulder X-rays, blood cultures, and trans-thoracic echocardiogram did not elucidate a source of infection. Non-contrast head computed tomography (CT) was normal. The rheumatology service was, thus, consulted and listed their primary differential diagnosis as polymyalgia rheumatica; however, they noted irreconcilability between this and his worsening infectious stigmata. This triggered further advanced imaging workup. A whole-body positron emission tomography/CT scan showed hypermetabolism of the right greater than the left shoulder, left hip joint, left thumb, and lateral wrist, suggestive of multifocal arthritis and myositis. Gadolinium-enhanced magnetic resonance imaging (MRI) brain demonstrated small foci of restricted diffusion in the subcortical left cerebellar and left posterior parietal lobes with corresponding T2/fluid-attenuated inversion recovery hyperintensity most consistent with subacute embolic infarcts. Enhanced MRI of his cervical spine, however, revealed C6–7 discitis/osteomyelitis and an associated 2.8 cm ventral SEA, as well as discitis/osteomyelitis of the C2 vertebral body and C5–6 endplates, for which the neurosurgery team was ultimately consulted [Figures 1 and 2].

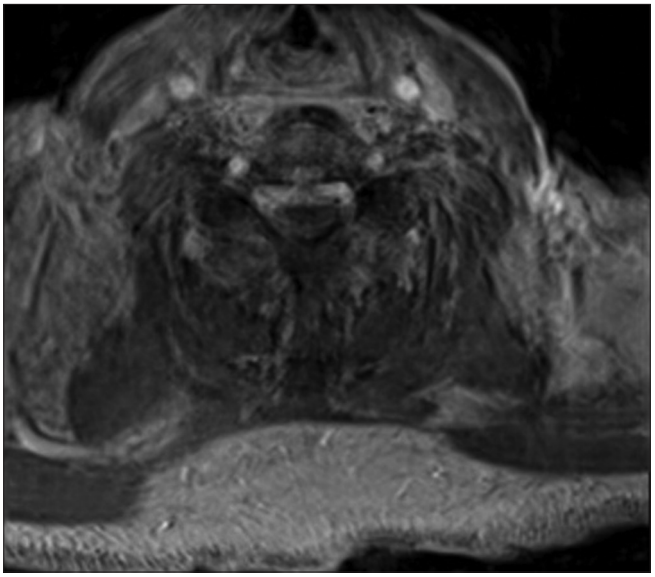

Figure 1: Motion degraded axial T1 post contrast image at the level of C6 demonstrating ventral epidural abscess leading to severe cord compression.

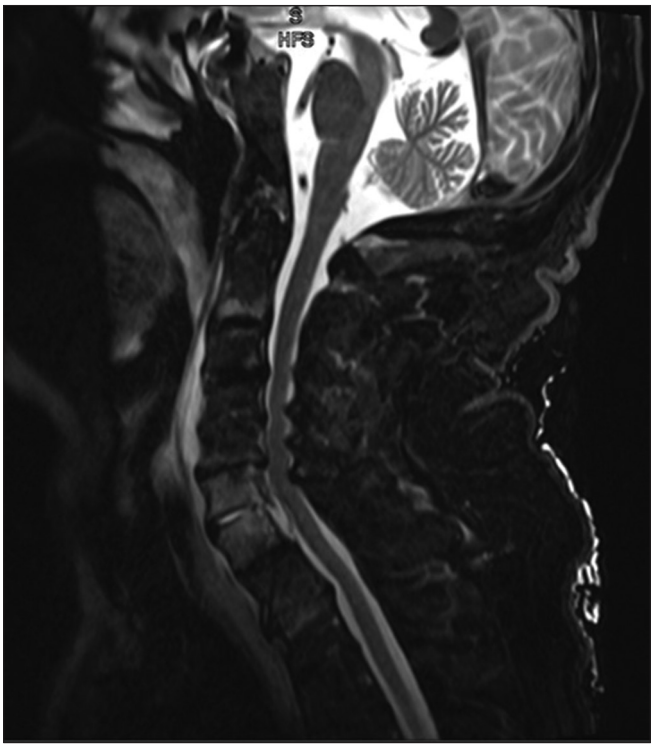

Figure 2: Sagittal T2 image demonstrating C6–7 discitis/osteomyelitis with associated prevertebral edema and ventral spinal epidural abscess. There is an abnormal signal noted in the C2, C5, C6, and C7 vertebral bodies.

Neurosurgical evaluation and intervention

On examination, the patient's mental status and cranial nerves were intact. He had pain-limited weakness in the bilateral deltoids, objective weakness in the distal left upper extremity and left lower extremity, and positive Babinski signs bilaterally. Given these clinical and corresponding radiographic findings, we recommended multilevel cervical laminectomy for decompression of his spinal cord and the acquisition of samples to guide antibiosis.

On hospital day 5, laminectomies of C3 through C7 were performed using the ultrasonic bone scalpel, as is our institutional norm. Intraoperative ultrasound confirmed adequate dorsal decompression despite the residual ventral collection. Shortly after laminectomy, the patient had improved

signals per intraoperative neuromonitoring in bilateral deltoid motor-evoked potentials, stable somatosensory-evoked potentials, and low amplitude improvement in the bilateral lower extremities. The "lobster-tailed" lamina was sent for culture; no definitive infection was identified. The spinal column was left un-instrumented, given the concern for active infection. A suprafascial wound vacuum was placed to allow for healing with the secondary intention of avoiding the high risk of subsequent wound breakdown.

Postoperative course

The patient returned to the ward where, over the coming days, he noted increased left deltoid and hip flexor strength. His blood was sent for Karius testing™ as recommended by the infectious disease team on hospital day 5. This testing yielded a result of *S. moniliformis* 1 week later, an organism associated with RBF. When this came to light and on further questioning, the patient noted that while he did not have any recollection of direct rat encounters, he was aware that one or more were occupying the vents at his place of employment. That same day, a transesophageal echocardiogram demonstrated centimeter-sized vegetations on the posterior mitral valve leaflet which was felt to be the likely source of the multifocal central nervous system emboli and seeding. The infectious disease team recommended 2 g of intravenous (IV) ceftriaxone daily for 6 weeks. The patient deferred cardiac intervention for the demonstrated vegetation. The patient was discharged home safely on hospital day 19. He was instructed to follow up in our neurosurgery clinic but was unfortunately lost to follow-up.

DISCUSSION

S. moniliformis is a highly pleomorphic bacterium, appearing as a filamentous, Gram-negative, nonmotile, and non-acid-fast rod.[4,18] It is the primary causative agent of RBF in North America.[19] "Sodoku" is another iteration of RBF caused by infection with *Spirillum minus,* which is more commonly seen in Asia.[5,7,19] Two distinct entities may result from *S. moniliformis* exposure through two distinct transmission modes. Contact with rats through bites, scratches, or excrement can lead to the classic RBF disease.[7,19] Alternatively, oral ingestion of *S. moniliformis* through contaminated food may lead to Haverhill fever, so named for a well-documented 1926 outbreak in Haverhill, Massachusetts.[3,5,10,19] Haverhill fever causes a constellation of symptoms that are identical to that of RBF, but it occurs without direct exposure to rats.[5]

Rats are considered the primary natural reservoir of *S. moniliformis*, but guinea pigs, gerbils, ferrets, and squirrels can also carry it.[1,4,19] Given a higher incidence rate of rate bites among low-income populations,[9] RBF has historically been associated with poverty.[5,13,14] Laboratory technicians are also affected by cases of RBF as a consequence of work-related exposures.[3,5,7] However, due to the increasing popularity of pet rats in recent years, there has been a concordant rise in RBF among children and pet store workers.[3,7,18] This is particularly concerning when considering that studies have found a nearly 10% transmission rate after being bitten, with a mortality rate of 13% if left untreated.[7,15,18,20]

RBF is a systemic illness that can present acutely or follow a relapsing remitting course.[3,4,18] A broad spectrum of nonspecific symptoms characterizes clinical presentations of RBF.[5,8] Affected individuals often experience relapsing fever, headache, vomiting, malaise, and migratory polyarthralgia.[4,5,7,15,20] A characteristic rash, erythematous and affecting the extremities, is also frequently observed.[19] If a rat bite causes infection, the bite is typically well-healed by the time of patient presentation without any residual inflammation.[5,7,18] Untreated RBF may progress to septic polyarthritis or lymphadenopathy and lead to various complications, including endocarditis, pericarditis, amnionitis, septicemia, interstitial pneumonia, prostatitis, pancreatitis, splenic or renal infarction, meningitis, and brain abscess.[1,4,5,15,18,19] Infections of the human spine are rarely reported but can manifest as spondylodiscitis, a psoas abscess, or an epidural abscess, as in this case.[2,8]

Given the nonspecific presentation of RBF and difficulties associated with the isolation and identification of *S. moniliformis*, diagnosis of the illness requires a high index of suspicion. Growth of *S. moniliformis* requires microaerophilic conditions and a specific culture medium enriched with 10–30% blood or serum.[4,20] However, most blood culture mediums contain sodium polyanethol sulfonate, an anticoagulant that inhibits the growth of *S. moniliformis* at concentrations >0.02%.[3,4,11,15] Several other assays offer increased sensitivity for detection, including gas-liquid chromatography, polymerase chain reaction, and 16S ribosomal ribonucleic acid (16S-rRNA) sequencing.[4,5,20] Advanced sequencing techniques such as meta-next-generation sequencing have been shown to enhance the speed and accuracy of detection significantly[20] The recommended course of treatment for RBF is IV penicillin G.[5,7] Doxycycline, tetracycline, cephalosporins, carbapenems, erythromycin, clindamycin, and ceftriaxone are also suitable substitutes.[3,20]

To date, there have only been two previous reports of patients with an epidural abscess caused by *S. moniliformis*; neither of these have involved the cervical spine. In 2012, a 58-year-old male presenting with a 2-week history of right-sided flank pain, fevers, and lower extremity weakness was found to have an abscess located between the L4 and S1 vertebrae.[2] The patient was diagnosed with RBF after subcultures of bacteria obtained from initial blood cultures of the abscess fluid

revealed *S. moniliformis*. Retrospective 16S-rRNA sequencing confirmed the presence of *S. moniliformis* in the abscess and the blood culture. As in this case, successful treatment with IV ceftriaxone led to complete resolution of the abscess. In 2017, a 40-year-old male complaining of diffuse abdominal pain was found to have an epidural abscess at the T6–8 vertebrae.[8] Initial diagnosis of RBF was made by 16S-rNA sequencing of samples taken from the abscess fluid. Cultures obtained from the abscess also underwent testing by matrix-assisted laser desorption ionization with time of flight, which confirmed the presence of *S. moniliformis*. The patient was successfully treated with penicillin G following diagnosis. Both patients had previously been exposed to rats, although the first patient's history of exposure was obtained retrospectively, while the second patient reported the recent occurrence of a rat bite before his diagnosis. Our case is the first reported to our knowledge to involve the cervical spine. RBF is rarely considered in the diagnosis of cervical SEA, especially in lieu of recent known exposure. However, our experience underscores the importance of maintaining a certain degree of suspicion for rarer, more fastidious organisms when seeking the source of a cervical SEA and likewise stresses the critical role of neurosurgical evacuation and simultaneous sample acquisition to prevent neurological decline and guide appropriate antibiosis.

Although SEA predominantly affects the lumbar or thoracic spine, epidural abscess in the cervical spine has been identified in 18–36% of cases.[12,16] Despite its low incidence, cervical SEA is often associated with more severe neurological deficits and an increased risk of morbidity and mortality.[12,17] Viable treatment options for cervical SEA include medical management and surgical intervention. Medical management is recommended for patients with significant comorbidities contraindicating surgery or those lacking neurological deficits.[17] Surgical intervention is indicated in cases of conservative treatment failure, persistent symptoms, neurological deficits, spinal instability, extended abscess (>2.5 cm), spinal deformities, or sepsis.[12] Surgical objectives include decompression of the epidural space, drainage of the abscess, restoration of spinal stability, and sampling of the infected tissue to guide antibiosis.[6,12,16,17] Postoperative antimicrobial therapy for 4–6 weeks is commonly prescribed to prevent recurrence of infection.[12] Notably, surgical intervention emerges as the preferred treatment approach in many cases.[12,17] Regardless of the treatment modality, prompt diagnosis and management of SEA are imperative to improve patient outcomes, as delays can lead to exacerbation of symptoms and increase the risk of mortality. In the present case, timely management and surgical intervention were critical in achieving a favorable patient outcome.

CONCLUSION

S. moniliformis is an incredibly uncommon pathological organism, especially as a cause of SEA. Our case is only the third in the neurosurgical body of literature to describe a patient with an epidural collection requiring neurosurgical decompression. Neurosurgeons should consider this rare entity in the differential diagnosis of SEA, and further representation in the neurosurgical literature may help improve early suspicion, identification, and surgical management of this morbid and potentially mortal disease. A cohesive multidisciplinary team of hospitalists, infectious disease experts, and neurosurgeons remains critical to optimizing outcomes in these rare and challenging cases.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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