

Spinal epidural abscess associated with an epidural catheter in a woman with complex regional pain syndrome and selective IgG3 deficiency

A case report

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Abstract

Rationale: Continuous epidural infusion of local anesthetic may be an alternative to sympathetic blocks in refractory cases of complex regional pain syndrome (CRPS). Spinal epidural abscess (SEA) is a well-known complication associated with this technique, especially in patients with immune deficiencies. We herewith report a cervical SEA associated with an epidural catheter in a woman with CRPS and selective IgG3 subclass deficiency.

Patient concerns: Severe pain interfering with activities of daily living.

Diagnosis: Complex regional pain syndrome type-1 with involvement of upper extremity.

Interventions: The patient underwent inpatient epidural infusion for management of left upper extremity CRPS. Her history was notable for previous left shoulder injury requiring numerous surgical revisions complicated by recurrent shoulder infections, and selective IgG3 deficiency. She received antibiotic prophylaxis and underwent placement of a C6–C7 epidural catheter. On day 5, she became febrile. Neurological examination remained unchanged and an MRI demonstrated acute fluid collection from C3–T1. The following day she developed left arm weakness and was taken for emergent cervical decompression. Intraoperative abscess cultures were positive for *Pseudomonas aeruginosa*.

Outcomes: Postoperatively, the patient's neurological symptoms and signs improved.

Lessons: Patients with selective IgG3 deficiency who are being considered for epidural catheterization may benefit from expert consultation with infectious diseases specialist. A history of recurrent device- or tissue-related infections should alert the clinician to the possible presence of a biofilm or dormant bacterial colonization. Close monitoring in an ICU setting during therapy is recommended. In case of early signs of infection, clinicians should have a high suspicion to rule out a SEA in immunocompromised patients.

Abbreviations: CRPS = complex regional pain syndrome, EMG = electromyography, ICU = intensive care unit, ID = infectious diseases, Ig = immunoglobulin, LUE = left upper extremity, MRI = magnetic resonance imaging, NSICU = neurosciences intensive care unit, PACU = post-anesthesia care unit, SEA = spinal epidural abscess, VAS = visual analog rating scale.

Keywords: abscess, catheter, complex regional pain syndrome, epidural, IgG3 deficiency

1. Introduction

Complex regional pain syndrome (CRPS) is a debilitating neurological condition that is characterized by a variable clinical

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course ranging from a mild and self-limited picture to a severe, disabling pain condition that interferes with every aspect of life.^[1] The primary approach to treating CRPS consists of conservative management with physical therapy and symptom-oriented pharmacologic therapy. In cases that fail to respond adequately to conventional therapy, interventional management may be considered, such as sympathetic blocks, neuraxial anesthesia, and neuromodulation.^[2] In refractory cases, continuous epidural infusion of local anesthetic has been shown to be an effective alternative to repeated injections.^[3–7] There are risks and complications, however, associated with this technique such as hypotension, spinal epidural abscess (SEA), hematoma, and meningitis.^[8] Patients that are especially at risk for infection are those with immune deficiencies.^[9]

The 4 subclasses of immunoglobulin (Ig) G differ from each other structurally and functionally by the nature of their constant (Fc) regions.^[10] IgG3 responds primarily to protein antigens and is an activator of the complement system; it also has an important role in antibody-mediated phagocytosis.^[11] Patients with selective IgG3 subclass deficiency have a propensity for infections and typically present with recurrent upper respiratory tract infections.

SEA in the setting of IgG3 deficiency has not previously been reported; however, a case of SEA associated with IgG4 deficiency has been reported.^[12] We herewith report a cervical epidural abscess associated with an epidural catheter in a woman with CRPS and selective IgG3 subclass deficiency.

2. Case report

The patient provided permission for publication of this case report. Hospital for Special Surgery institutional review board approval was obtained for this case study. A 29-year-old woman presented for inpatient continuous epidural infusion for management of worsening CRPS of the left upper extremity (LUE). The patient's history began 3 years ago with a sports-related injury to her left shoulder that required over a dozen surgical revisions, some of which were complicated by recurrent and prolonged shoulder infections. Several different microorganisms had been isolated during the various episodes of infection, including *Pseudomonas aeruginosa*, *Sphingomonas paucimobilis*, *Candida colliculosa*, and *Staphylococcus aureus*. The patient's most recent surgery was a left shoulder tendon release and revision performed ten months ago, which was followed by the development of CRPS. Her symptoms included severe pain, allodynia, edema, muscle spasms, and temperature changes of her LUE. An electromyography (EMG) evaluation by a neurologist revealed a brachial plexus injury. Her past medical history was also notable for asthma and selective IgG3 deficiency diagnosed 2 years prior during work up for recurrent joint infections. The patient's CRPS symptoms were refractory to oral medical management, which included opioids, antidepressants, and antispasmodics. She had significant, although short-lived, symptom relief with left stellate ganglion blockade. Given her favorable response to sympathectomy, she had 2 prior admissions for continuous cervical epidural infusions that provided 4 to 6 weeks of decreased pain, improved participation with physical therapy, and a reduction in oral opioid requirements.

The patient underwent placement of a left C6–C7 interlaminar epidural catheter under fluoroscopic guidance (Fig. 1) and was then transferred to the postanesthesia care unit (PACU) for continuous monitoring. Preprocedure labs including a complete blood count, complete metabolic panel, and creatinine

phosphokinase were normal. Given her history of selective IgG subclass deficiency and atypical infections in the past, she received antibiotic prophylaxis with 1 gram of intravenous vancomycin one hour prior to the procedure. Following an uneventful placement, the catheter was ruled out for intrathecal and intravascular placement with 3 mL of 1% lidocaine with 1:200,000 epinephrine. Thereafter, an epidural infusion of 0.25% bupivacaine with hydromorphone 10 mcg per mL and clonidine 1 mcg per mL was started at a continuous rate of 4 mL per hour. Oral home medications were also continued including methadone 10 mg twice a day, hydromorphone 4 to 8 mg as needed, diazepam 10 mg 4 times a day, baclofen 20 mg 4 times a day, and amitriptyline 100 mg once a day. The patient tolerated the therapy well and reported a decrease in pain from 9/10 to 7/10 on the visual analog rating scale (VAS) as well as less muscle spasms.

On hospital day 2, the continuous infusion was increased to 6 mL per hour and a demand dose of 2 mL every 15 minutes was started. The patient continued to do well, noting improved sleep and a further decrease in LUE spasms and edema, as well as, a VAS of 5/10. Prophylactic vancomycin was continued daily and dosed according to trough levels. At 9:00 PM on hospital day 5, the patient became febrile to 38.1°C and the decision was made to wean the infusion more rapidly than originally planned and to remove the epidural catheter as soon as possible. This was achieved by 2:00 AM on hospital day 6.

A few hours later, the patient developed progressive headache, neck pain, and a further increase in temperature to 40.0°C; however, neurological examination at this time remained unchanged from her baseline. The epidural site was nontender to palpation and did not exhibit any signs of active infection. Blood and urine cultures were sent, a chest x-ray was negative for an active pulmonary process, and laboratory workup was notable for an increase in white count to $9.7 \times 10^3/\mu\text{L}$ from a day of admission value of $6.0 \times 10^3/\mu\text{L}$. Per infectious disease (ID) recommendations, a cephalosporin (cefepime) was added empirically to vancomycin, which the patient responded to favorably with abatement of fever and a decrease in white count to $5.9 \times 10^3/\mu\text{L}$. At 11:30 AM the patient underwent an MRI of the cervical spine utilizing sagittal T1- and T2-weighted imaging,

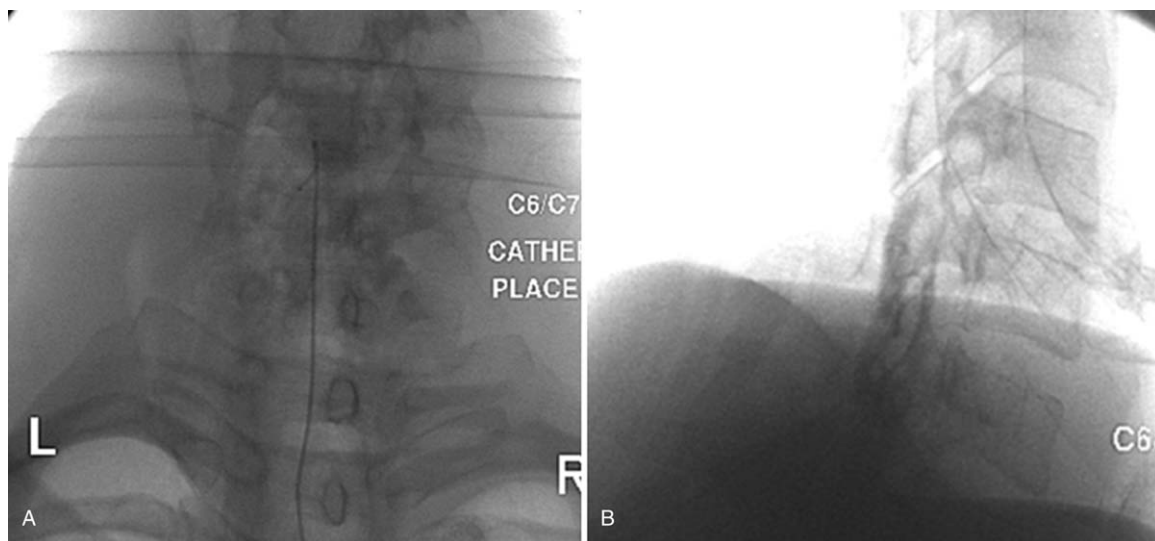


Figure 1. Procedural fluoroscopy. (A) AP and (B) lateral views of epidural catheter placed in the left C6–C7 interlaminar space with distal tip in the C4–C5 interspace.

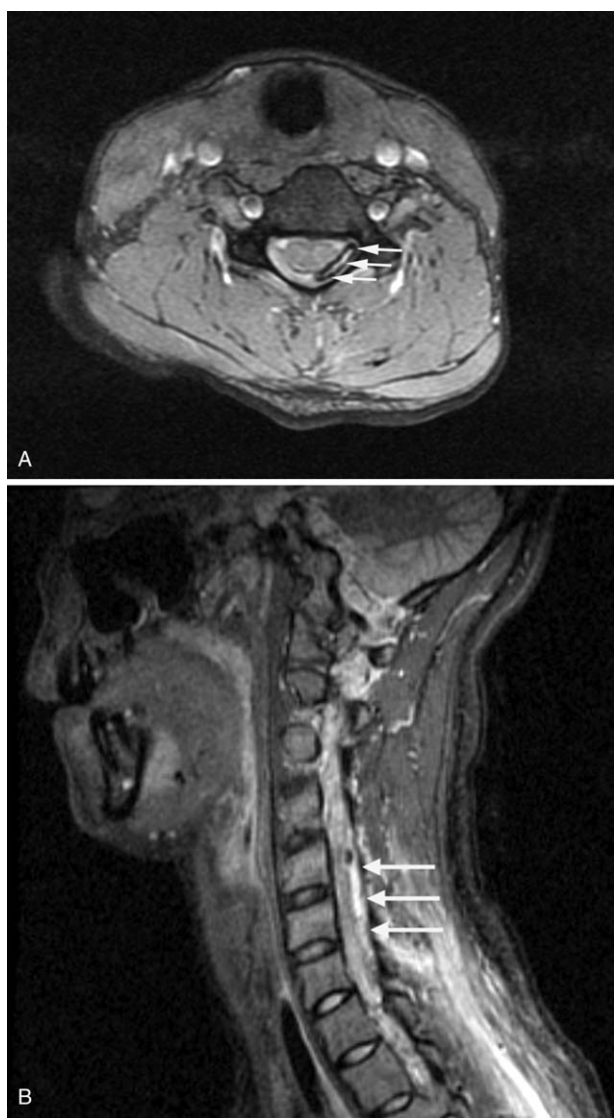


Figure 2. Cervical spine MRI with IV contrast. (A) Axial 2D multiple echo recombined gradient echo (MERGE) view and (B) sagittal inversion recovery (IR) view showing a fluid collection in the cervical epidural space, coursing from C3 to T1 (white arrows). MRI=magnetic resonance imaging, MERGE=multiple echo recombined gradient echo.

sagittal inversion recovery, axial fast spin echo and gradient recall techniques. Imaging demonstrated an epidural collection from the inferior margin of C3 to T1 compressing the left C5 and C6 nerve roots with some effacement of the thecal sac but no cord deformity as well as considerable interstitial edema in the left paraspinal muscles (Fig. 2).

Given the clinical picture and the patient's complicated infectious history, after neurology consultation, the decision was made to transfer her to the neurosciences intensive care unit (NSICU) for further management of suspected epidural abscess. The patient underwent hourly neurological examination, which remained normal until 24 hours later when she developed intractable nausea and vomiting, and left arm weakness. She was taken to the operating room for emergent decompression and evacuation via C3 to C7 cervical laminectomies and C4–C5 and C5–C6 left foraminotomies. Intraoperatively, the epidural space

was noted to be filled with a large inflammatory mass compressing the spinal cord as well as several pockets of purulent material. Intraoperative cultures of the cervical abscess were positive for *P aeruginosa* susceptible to cefepime. Urine and blood cultures from hospital day 6, however, were negative. Vancomycin was stopped and cefepime was continued per ID recommendations. The patient had resolution of arm weakness and an uneventful postoperative course. She was discharged home on postoperative day 3 where she continued intravenous cefepime for a total of 6 weeks.

3. Discussion

To the best of our knowledge, this is the first case report of an epidural catheter complicated by SEA in a patient with selective IgG3 subclass deficiency. SEA is a relatively rare condition with an incidence of approximately 2 to 25 patients per 100,000 admissions per year.^[13] It is a neurosurgical emergency that often presents as a pyogenic infection localized between the dura mater and the vertebral periosteum within the spinal epidural space.

Patients who develop SEA are frequently immunocompromised and diabetes mellitus is the most common risk factor cited in studies.^[9] Other major predisposing risk factors for infection include old age, intravenous drug use, alcohol abuse, chronic renal insufficiency, malignancy, and steroid therapy.^[8,9,14] Infections near and distant from the vertebral column (e.g., skin abscess, vertebral osteomyelitis/discitis), trauma, and degenerative spinal disorders are important risk factors.^[8,14] Invasive procedures, mainly epidural anesthesia, extraspinal and spinal operations, and vascular access, led to 22% of SEA cases in the literature.^[14] In the case of a difficult epidural catheter insertion, multiple passes of the needle may create a subcutaneous hematoma that goes on to act as a nidus for infection.^[8] Bacteria may also reach the epidural space by hematogenous spread or contiguous extension from neighboring organ structures.^[8,14] In many cases, multiple risk factors and potential infection sources may be present in individual patients.

Our patient underwent an uneventful, single pass placement of an epidural catheter under sterile technique and fluoroscopic guidance. She did not have any signs or symptoms of localized or systemic infection in the preprocedural period. Similar to her previous treatments, she received prophylactic antibiotics given her history of IgG3 deficiency and recurrent shoulder joint infections. Although gram-positive cocci, namely *S aureus*, are the most common organisms encountered in SEA formation and in bacterial septic arthritis,^[8,15,16] *P aeruginosa* was isolated from our patient's abscess. This pathogen is an uncommon infecting organism in epidural abscesses and atraumatic joint infections^[8,16]; however, it was also previously identified twice during our patient's recurrent shoulder infections.

Up to 20% of cases of septic arthritis may be caused by gram-negative bacteria, most commonly *Escherichia coli*.^[16] Comorbid conditions such as immunosuppression, chronic disease, intravenous drug abuse, chronic wounds, and arthritic joints predispose patients to gram-negative joint infections.^[16] *P aeruginosa* is an uncommon cause of septic arthritis, but is well-known to establish biofilms that lead to chronic tissue-related or device-related infections that are difficult to treat.^[16,17] Biofilm recalcitrance toward antibiotics is mostly related to their low growth rate ("dormancy"), the presence of highly tolerant bacterial subpopulations ("persisters"), and a microenvironment within the biofilm matrix that impairs antimicrobial activity.^[17,18] We believe that in our patient's case, inoculation of the

cervical epidural space occurred either hematogenously or by local spread of a dormant shoulder infection.

Patients with selective IgG3 deficiency who are being considered for epidural catheterization may benefit from expert consultation with ID. A history of recurrent device- or tissue-related infections should alert the clinician to the possible presence of a biofilm or dormant bacterial colonization. Those patients who are deemed high risk for SEA formation are obviously precluded from undergoing the intervention. Patients who undergo the procedure should have close monitoring in an intensive care unit setting during epidural infusion therapy. As presented in this case, neurological findings may be delayed in an epidural abscess. Therefore, in case of early clinical and laboratory signs of infection, clinicians should have a high suspicion to rule out a SEA in immunocompromised patients. Early imaging using MRI with intravenous contrast (or computed tomography scan, if MRI is contraindicated) along with routine infectious work up are recommended.

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