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Case Report

Compressive spinal epidural mass caused by *Propionibacterium acnes*

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Abstract

BACKGROUND CONTEXT: *Propionibacterium acnes* is a gram-positive and facultative anaerobe bacillus that is found within sebaceous follicles of the human skin and recognized as a cause of infections after spinal surgery. To our knowledge, there has been no previously reported case of symptomatic compressive chronic inflammatory epidural mass caused by *P. acnes* in a patient with no prior spinal procedures.

PURPOSE: This study aimed to describe a case of primary spinal infection by *P. acnes*.

STUDY DESIGN: This study is a case report of a condition not previously described in the literature.

METHODS: We present the history, physical examination, laboratory, radiographic, and histopathologic findings of a chronic inflammatory epidural mass caused by *P. acnes* in an immunocompetent adult male with no history of spinal surgery.

RESULTS: A 51-year-old man presented to our clinic with sudden onset bilateral lower extremity weakness, inability to ambulate, and urinary retention. His past clinical history was remarkable only for hernia and left knee surgery but no spinal surgery. A year earlier, he had an infected draining abscess of the right axilla that was successfully managed medically. At presentation, his serum erythrocyte sedimentation rate and C-reactive protein were moderately elevated. Pan-spine magnetic resonance imaging was notable for a circumferential epidural mass from C5 to T6. He underwent emergent decompression; the mass was removed and sent for culture and pathologic evaluation. Cultures from all three specimens collected during surgery grew *P. acnes*, and the patient was successfully managed on intravenous ceftriaxone, while pathology revealed a chronic inflammatory reactive process.

CONCLUSIONS: This is the first reported case of a primary spinal mass with chronic inflammatory features caused by *P. acnes*. In cases of epidural mass of unknown origin, both pathologic specimens and cultures should be obtained as slow-growing organisms may mimic oncologic processes.

Keywords: Compression; Epidural abscess; Laminectomy; Primary spinal infection; *Propionibacterium acnes*; Spinal cord

Introduction

Propionibacterium acnes is a gram-positive and facultative anaerobe bacillus found within sebaceous follicles [1]. Along with coagulase-negative staphylococcus species, it is one of the most common bacterial causes of infections after

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spinal surgery, particularly spinal fusions [2,3]. *P. acnes* has also been isolated from the intervertebral disc material of patients with severe sciatica endocarditis, various cerebrospinal fluid shunt infections, and prosthetic joint infections [2,4–6]. Because of its low virulence, patients with *P. acnes* infections may present months or years postoperatively with back pain or drainage around the incision site [7,8]. Infections occurring more than a decade after surgery have been reported [9–11].

To our knowledge, all previous reports of *P. acnes* infections of the spine were associated with a prior spine surgery. Here, we report a case of a symptomatic compressive chronic inflammatory epidural mass caused by this organism in an immunocompetent patient without a history of prior spinal intervention.

Case report

A 51-year-old man presented to our institution with back pain, acute bilateral lower extremity weakness and numbness, inability to ambulate, and urinary retention. The patient denied fevers, recent infections, and any trauma. He had a remote history of left knee surgery and hernia repair but no spinal interventions. He also had a history of an infected draining cyst of the right axilla that was managed medically 1 year before presentation. He had a 15-pack-year smoking history but had quit 4 years ago.

On physical examination, the patient was morbidly obese (BMI 44.3) and afebrile. He was tender over his low cervical and upper thoracic spine. The patient's upper extremities were neurologically intact, but he had 1/5 strength in his hip flexors, 2/5 in the quadriceps and hamstrings, and 3/5 in his distal lower extremity motor groups. He had decreased sensation below T7, symmetric hyperreflexia in the lower extremities, and three beats of clonus bilaterally. Post-void residual was 1,400 mL; rectal tone was normal.

Although his CBC indices were within their reference ranges, the patient had moderately elevated erythrocyte sedimentation rate (42 mm/h) and C-reactive protein (CRP, 35.38 mg/L). Magnetic resonance imaging (MRI) identified a circumferential compressive lobulated epidural mass, T1 and T2 isointense with diffuse enhancement, extending from C5 to T6 with evidence of cord compression (Fig. 1, Left). Spinal cord edema was present at C6/C7, T1/T2, and T3/T4. Given the epidural mass and possibility of metastatic lesion, computed tomography scans of brain, chest, abdomen, and pelvis were performed to screen for additional lesions, but none was identified.

An emergent C7–T7 laminectomy and excision of the mass were performed within 4 hours after arrival at our institution. Thickened dural tissue and ligamentum flavum extending from C7 to T7 were noted intraoperatively. There was no frank purulence. Biopsies of the firm and fibrous thoracic epidural mass and dural inflammation were sent for gram stain, culture, and pathology. No instrumentation was performed because all facet joints were preserved and the anterior spinal elements were not affected. Postoperatively, the patient's lower extremity strength and sensation progressively improved. The patient was discharged to an inpatient rehabilitation facility on postoperative day 6, at which time he had 4/5 strength

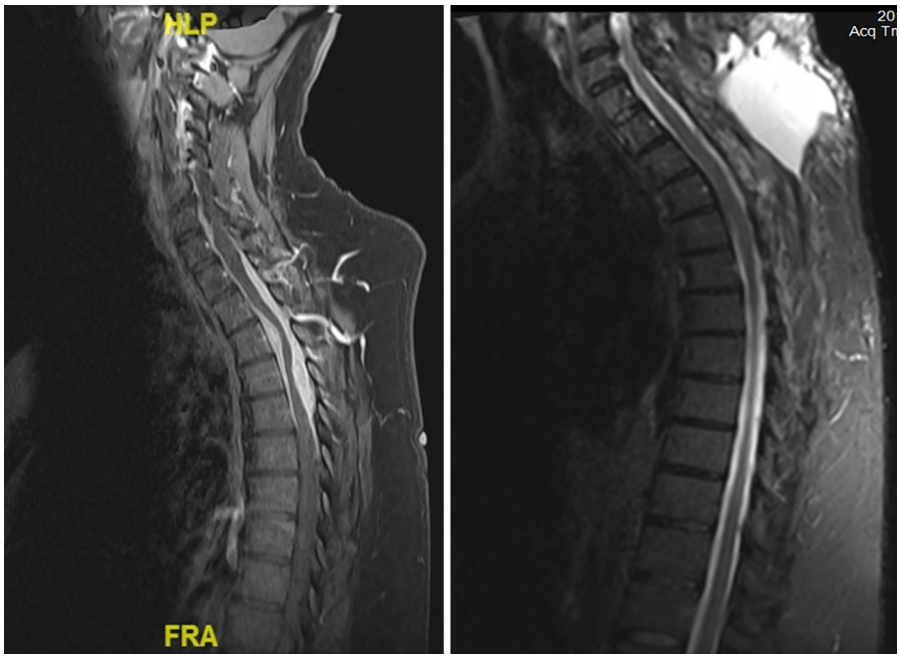

Fig. 1. (Left) Gadolinium-enhanced T1-weighted sagittal magnetic resonance imaging (MRI) at presentation with lobulated epidural mass extending from C5 to T6 with evidence of circumferential spinal cord compression. (Right) T2-weighted MRI at 6 weeks following decompressive laminectomy with resolution of spinal cord compression.

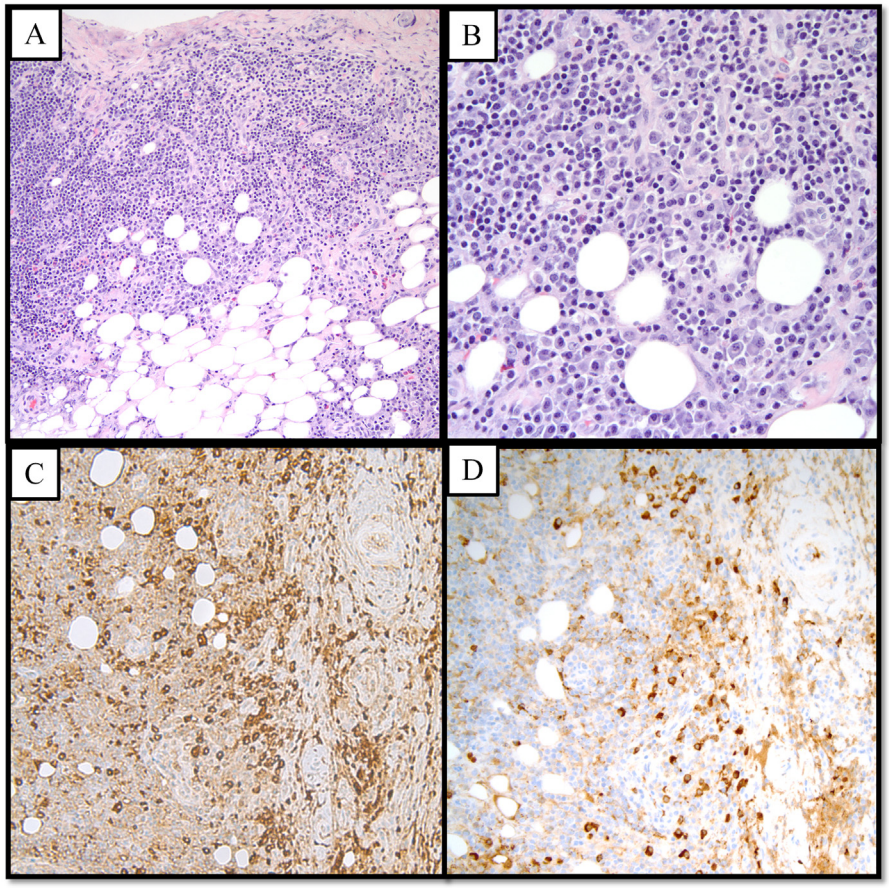

Fig. 2. Histologic examination revealed a dense lymphoplasmacytic population infiltrating fibroadipose tissue. (A) Hematoxylin and eosin, 100×. (B) Hematoxylin and eosin, 500×. (C) Anti-kappa antibody, 200×. (D) Anti-lambda antibody, 200×.

throughout his lower extremities and regained the ability to ambulate.

Nine days after surgery, all three cultures sent for examination grew *P. acnes*, and IV ceftriaxone was initiated. The erythrocyte sedimentation rate and CRP decreased to 28 and 29, respectively, by 2 weeks postoperatively. On histologic examination, the thoracic epidural mass consisted of a dense lymphoplasmacytic population with scattered eosinophils and rare mitoses (Fig. 2, A and B), and occasional formation of secondary follicles. By immunohistochemistry, the lymphoid population was comprised mostly of T-lymphocytes with rare secondary CD20 positive B-lymphoid follicles seen. The large MUM1+, CD138+plasma cell population consisted of a larger subset of kappa-positive than lambda-positive plasma cells, with an overall kappa to lambda ratio of 3–4:1 and in some areas reaching 5:1 (Fig. 2, C and D). These findings were also noted in the in situ hybridization studies. Molecular studies were reported negative for immunoglobulin heavy chain gene rearrangements and also negative for T-cell receptor beta and gamma gene rearrangements, and the fluorescence in situ hybridization study was negative for IgH (14q32.3) rearrangement, findings indicative of a reactive inflammatory process.

The patient continued to improve clinically with physical therapy and was discharged home on postoperative day 16 with plans for 6 weeks of ceftriaxone treatment and close follow-up with repeat MRI. At 6 weeks follow-up, his lower extremity strength and urinary and bowel function returned to normal with resolution of spinal cord compression on MRI (Fig. 1, Right).

Discussion

Previously thought to be merely a contaminant, *P. acnes* is now well recognized as an important pathogen in implant-associated infections. *P. acnes* has been implicated in infections of prosthetic joints, cardiac devices, breast implants, intraocular lenses, neurosurgical shunts, and spinal instrumentation [3]. It accounts for approximately 10% of prosthetic joint infections, with an increased incidence in the shoulder and spine, likely secondary to a higher concentration of sebaceous follicles at those anatomic sites [3].

In the pediatric and adolescent scoliosis literature, *P. acnes* is a well-known cause of postoperative complications of spinal fusion with instrumentation. Reported postoperative infection rates in pediatric and adolescent patients with scoliosis

vary between 0.5% and 14% [7,12,13]. In a recent case-control study involving 20 pediatric patients with deep surgical site infections after scoliosis surgery and 50 patients who did not, coagulase-negative *Staphylococcus* was isolated in eight patients and *P. acnes* in four patients [12]. Increased pre- and postoperative Cobb angles, non-ambulatory status, and an increased length of hospital stay were associated with an increased risk of infection [12]. Allograft bone use [7], higher volume of instrumentation [7], back acne [13], surgery duration greater than 6 hours [13], and stainless-steel alloy [14] have also been implicated as possible risk factors.

Diagnosis of *P. acnes* infection is challenging because of the fact that culture results must be interpreted with caution. Because of its slow growth, *P. acnes* should be allowed to grow for 14 days in aerobic and anaerobic agar as well as thioglycollate broth [3,15–17]. Additionally, biofilm formation further complicates detection and treatment [18,19], and *P. acnes* infection should be considered in cases of delayed infections and low-grade implant-associated infections even when cultures are negative [3]. Vortexing and sonication of explanted material has recently been shown to reliably increase yield by detaching the microorganism from its biofilm on implants of the spine [18], hip and knee [19], and shoulder [20].

If *P. acnes* grows from only a minority of tissue or fluid samples, other criteria for diagnosis such as clinical symptoms and signs, histopathology, and molecular tests should be sought as it may also grow as a contaminant [2]. One retrospective study suggested using the results of at least four deep sample cultures, histology, and CRP values to make the diagnosis [21]. Newer molecular methods may be able to diagnose infection even in cases of prior antibiotic administration.

In our case, the patient had no history of spinal intervention, which made suspicion for a *P. acnes* infection low. It is possible that the patient's axillary cyst infection, although treated 1 year before presentation, may have been the source of the epidural lymphoplasmacytic population with mass-like presentation. Following initial pathologic examination, a possible early epidural involvement by a low-grade B-cell lymphoma was also raised in the differential diagnosis. Primary spinal epidural lymphomas (PSELs) are rare entities, comprising only a small subset of all epidural masses [22]. The largest case series to date consisted of 52 patients treated at nine different institutions of the Rare Cancer Network over a 20-year period [23]. The patients in this series were predominantly male (69%) with a median age of 61 years and had disease of the thoracic spine (65%) [23]. Once an epidural lymphoma is diagnosed by MRI and tissue biopsy, a comprehensive systemic workup, including whole-body computed tomography, bone marrow biopsy, bone scintigraphy, and cerebrospinal fluid examination is recommended to evaluate the extent of the disease [22]. Of interest, a subset of marginal zone lymphomas is associated with an infectious etiology and some patients achieved complete remission only with antibiotic therapy [24].

In our patient, the atypical lymphoplasmacytic population lacked clonality by PCR and fluorescence in situ hybridization studies. In addition, three biopsy samples collected during surgery grew *P. acnes* in 2 weeks' interval. The case re-emphasizes that in patients with suspected malignancy of the epidural space, separate biopsy samples collected in sterile conditions during surgery should be submitted for microbiologic examination in an effort to increase the diagnostic accuracy. Also, detailed histologic examination with associated ancillary studies is needed to exclude a diagnosis of lymphoma.

Conclusions

P. acnes has been previously been reported to cause spinal infections following spinal intervention, but it is a previously unreported cause of idiopathic epidural infection. We report a case of a chronic inflammatory epidural mass caused by *P. acnes* that clinically mimicked lymphoma in an adult male without a prior history of spinal surgery. Early detection of the microorganism and a specific antibiotic therapy alleviated his symptoms and helped with cure. Our case draws attention to the complex presentation of an infectious process and to the importance of ancillary studies in reaching the diagnosis.

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