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Case Report

Isolated cervical *Cutibacterium acnes* osteomyelitis in a patient with no primary source of infection – A case report and review of the literature

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Received: 27 June 2023
Accepted: 18 September 2023
Published: 06 October 2023

DOI
10.25259/SNI_542_2023

Quick Response Code:

ABSTRACT

Background: Cervical vertebral osteomyelitis (CVO) is a rare pathology that leads to progressive osseous degradation and eventual loss of bone putting the patient at risk of devastating neurological injury in the event of bony collapse or instability. *Cutibacterium acnes* formerly called *Propionibacterium acnes* is rare, but within the last two decades has been an increasingly reported cause of osteomyelitis. The majority of *C. acnes* vertebral osteomyelitis cases have been reported in patients with a history of prior invasive procedures where direct contamination at the time of procedure was suspected as the underlying etiology.

Case Description: We report a unique case of an otherwise healthy 39-year-old male with no prior history of invasive procedures who presented with CVO secondary to *C. acnes.* He underwent surgical debridement and fusion in conjunction with antibiotic treatment. The patient recovered well and a 2-year follow-up with serial imaging showed no evidence of disease recurrence.

Conclusion: *C. acnes* is an under-recognized and under-reported etiology of spine infections. Clinicians should be aware of the pathological potential and atypical presentation of *C. acnes* vertebral osteomyelitis.

Keywords: Cervical vertebral osteomyelitis, *Cutibacterium acnes*, Spine surgery

INTRODUCTION

Cervical vertebral osteomyelitis (CVO) is a rare pathology that leads to progressive osseous degradation and eventual loss of the infected neck bone.[6,13] The large diameter of the cervical spinal cord relative to the spinal canal – in conjunction with a highly mobile neck – puts the patient at risk of devastating neurological injury in the event of bony collapse or instability.[4]

CVO has a reported incidence of about 3–11% of all osteomyelitis cases involving the spine.[2,13,16] This affliction tends to present predominantly in male older adults usually around the age of 57–60 years.[2] Predisposing comorbidities include diabetes, tuberculosis (TB), liver disease, prior surgical history, alcoholism, drug abuse, and dental work.[2,4] The frequently identified microbiological culprits in patients with osteomyelitis are opportunistic gram-positive staphylococci (75% of cases),[18] the most common of which is *Staphylococcus aureus*.[14] This is

due to its high prevalence (20% of the population)[11] in skin microflora and mucosal surfaces.[6] *S. aureus* may invade through extension from another local site of infection, hematogenous spread, or through trauma resulting in direct seeding.[16]

Cutibacterium acnes formerly called *Propionibacterium acnes* is a nonspore-forming anaerobic Gram-positive bacillus that is also found in skin microflora[9] and is a rare, but increasingly reported, cause of osteomyelitis.[17] Within the last two decades, there has been an observed rise in the number of pyogenic vertebral osteomyelitis infections, along with cases caused by *C. acnes*, correlated with the rising number of patients undergoing spinal surgeries as well as pain injections to the epidural and paraspinal regions.[8] The majority of *C. acnes* vertebral osteomyelitis cases have been reported in patients with a history of prior invasive procedures where direct contamination at the time of procedure was suspected as the underlying etiology.[8]

In this article, we report a unique case of an otherwise healthy 39-year-old male with no prior history of invasive procedures who presented with CVO secondary to *C. acnes*.

CASE PRESENTATION

A 39-year-old male with no previous history of invasive procedures or primary *C. acnes* infection presented to the emergency department 1 week after the insidious onset of severe posterior neck and back pain, muscle spasms, and bilateral hand numbness. The patient was afebrile but initial labs were remarkable for mildly elevated leukocytes (12.26×10^9/L) and significantly elevated C-reactive protein (CRP) (7.8 mg/L). The patient was started on cefazolin after cultures were drawn. A computed tomography (CT) scan showed comminuted compression deformity of the C6 vertebral body with 50–55% height loss and retropulsion of bony fragments into the spinal canal resulting in moderate thecal sac narrowing. There was no underlying lytic lesion to suggest a neoplastic process and no gross paraspinal soft-tissue abnormality was noted [Figure 1].

On magnetic resonance imaging (MRI), there was a redemonstration of a comminuted C6 compression fracture with retropulsion of bone fragments into the spinal canal resulting in a thecal sac anterior-posterior width of 7 mm with patency of the bilateral neural foramina [Figure 2]. Postcontrast T1 sequence MRI showed C6 vertebral body and bilateral pedicle enhancement with C2-T1 prevertebral soft-tissue swelling of variable thickness (maximum width of 6 mm). The paraspinal soft tissues were again otherwise unremarkable [Figure 3]. MRI T2 sequence showed regional flow voids within the C6 vertebral body [Figure 4]. There was diffusion restriction on the apparent diffusion coefficient and diffusion-weighted imaging diffusion restriction (not shown).

The patient underwent a C6 corpectomy followed by C5-C7 anterior cervical fixation. The patient tolerated the procedure well and the post-operative physical exam was at baseline. Intraoperative cultures and polymerase chain reaction confirmed the diagnosis of *C. acnes* osteomyelitis. The patient was discharged with a 14-day course of Cefalexin.

The patient was subjected to a comprehensive monitoring period spanning 2 years, during which both CT and MRI examinations consistently revealed the absence of any recurring infection with stable postsurgical changes related to C6 corpectomy and C5-C7 anterior fixation. These findings serve as evidence of successful antibiotic treatment and eradication of the infectious microbe. In addition, the patient has fully recovered their physical strength and

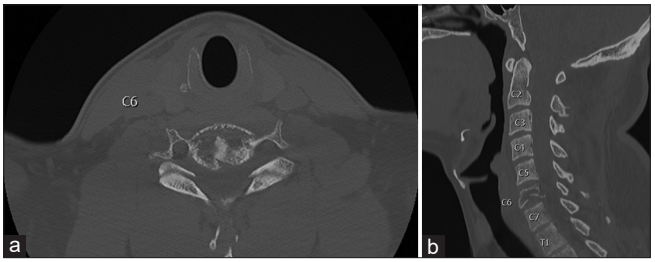

Figure 1: Preop CT axial (a) and sagittal (b) view showing comminuted compression deformity of C6 vertebral body with 50-55% height loss and retropulsion of bony fragments into the spinal canal.

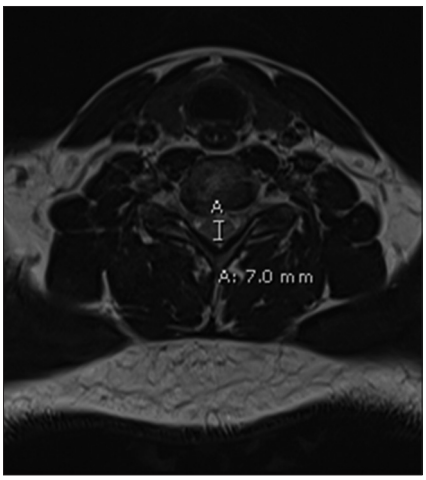

Figure 2: Preop MRI showing redemonstration of a comminuted C6 compression fracture with retropulsion of bone fragments into the spinal canal resulting in a thecal sac anterior-posterior (AP) width of 7 mm with patency of the bilateral neural foramina.

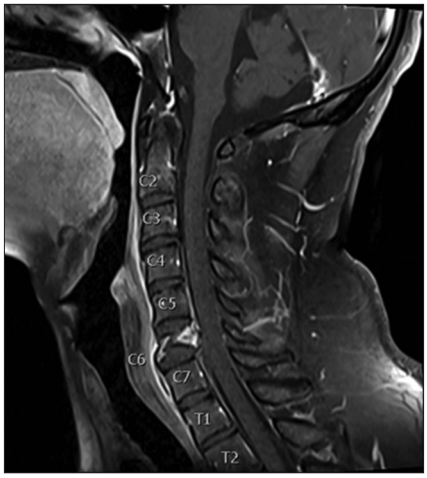

Figure 3: Post-contrast T1 sequence MRI showed C6 vertebral body and bilateral pedicle enhancement with C2-T1 pre-vertebral soft tissue swelling of variable thickness (maximum width of 6 mm). The paraspinal soft tissues were again otherwise unremarkable.

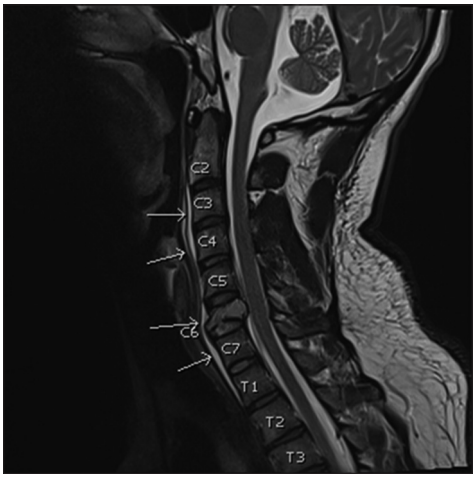

Figure 4: MRI T2 sequence showed regional flow voids within the C6 vertebral body.

sensation in all limbs, and any previously experienced pain has completely resolved.

DISCUSSION

While *C. acnes* vertebral osteomyelitis appears to occur almost exclusively in patients who have undergone prior invasive procedures,[7] there are a scarce number of reported cases in the literature where *C. acnes* vertebral osteomyelitis occurred in patients with no history of prior surgeries.[10,16] In a previously published case series of *C. acnes* vertebral osteomyelitis, six out of the nine patients with *C. acnes* vertebral osteomyelitis underwent prior surgeries to the vertebral column (4 with laminectomies and 2 with discectomies).[8] Another report described the case of a 27-year-old male with Reiter's syndrome who had steroid-induced *C. acnes* infection that seeded the bone causing C4-C7 osteomyelitis.[14] There was also a reported case of an elderly male who initially presented with neck pain and elevated ESR with inflammation noted at C5-C6. The patient underwent needle aspiration that only grew a scant amount of *C. acnes* raising the concern that the isolated *C. acnes* was a skin contaminant.[10] Carragee reported a case series of 111 patients with 41 having native, non-implant-related spinal osteomyelitis secondary to low-virulence organisms, 4% of which were due to *C. acnes*.[3] A novel case of *Cutibacterium modestum*, a *C. acnes* mimic, was recently reported in an 82-year-old man presenting with fever and exacerbation of the previous L4 lumbago. He was treated with ampicillin followed by amoxicillin.[15] *C. modestum* is very similar to *C. acnes* in its microbiological characteristics but is also a difficult-to-identify organism requiring a combination of matrix-assisted laser desorption ionization, microbial biochemical analysis, and gene sequencing to correctly recognize.[15]

There are several unique attributes of *C. acnes* vertebral osteomyelitis as compared to other pyogenic causes of vertebral osteomyelitis. The first is that while most patients diagnosed with *C. acnes* vertebral osteomyelitis initially present with the nonspecific complaint of back pain, these symptoms are rarely accompanied by fever, elevated CRP, and ESR, and almost always have a history of a previous spine procedure.[7,8] Imaging is also usually indeterminate and can present similarly to degenerative changes, necessitating tissue culture. This contrasts with other common causes of vertebral osteomyelitis that can be sufficiently diagnosed with imaging.[7] While this could be due to a lower virulence as compared to other causes of pyogenic osteomyelitis like *S. aureus*,[8] it could also be because the average duration of time between the onset of symptoms and diagnosis between *C. acnes* and other causes of pyogenic vertebral osteomyelitis is much longer. For example, the average duration of time between symptom onset and diagnosis for *C. acnes* vertebral osteomyelitis is around 4.7 months. Comparatively, it is closer to 2 months for TB and 3.7 weeks for *S. aureus*.[16] Another interesting element observed in radiographic studies of patients with *C. acnes* vertebral osteomyelitis – and in this case – is the notable lack of significant paravertebral and epidural inflammation. This again could be related to the lower virulence of *C. acnes* as compared to other pyogenic causes of vertebral osteomyelitis.

There is no current standardized, evidence-based guideline for the management of *C. acnes* CVO.[12] In general, medical

treatment of osteomyelitis involves tailored intravenous antibiotic therapy for 6–8 weeks followed by another 6-week course of orally administered antibiotics until the infectious agent is eliminated. Treatment with antibiotics for <4 weeks has been reported to have a relapse rate of 25%. However, the best type of antibiotic and appropriate length of time for antibiotic administration are still unknown and need further studies.[1,4,5,16,17] *C. acnes* is generally susceptible to doxycycline, rifampin, fluoroquinolones, clindamycin, metronidazole, and beta-lactams.[12]

Passerini *et al*. conducted a study on nonhardware-associated vertebral osteomyelitis, examining its characteristics and outcomes.[12] They discovered that most patients had lesions affecting the thoracic spine, with a preceding event at the infection site. The researchers proposed that *C. acnes* should be included in the microbiological differential diagnosis for individuals suspected of having vertebral osteomyelitis, even though it is sometimes regarded as a contaminant. According to their findings, *C. acnes* exhibited initial growth at or after 7 days of anaerobic incubation, indicating its slow-growing nature. Therefore, they recommend incubating anaerobic cultures for 14 days to detect the growth of *C. acnes* in suspected culture samples. Notably, the time taken for patients with *C. acnes* considered a contaminant to show positive results was longer compared to patients with *C. acnes* vertebral osteomyelitis.[12]

CONCLUSION

The laboratory and radiographic findings of *C. acnes* nonimplant-associated vertebral osteomyelitis are not well-characterized. However, what is known is that they often present with atypical findings. Due to the slow-growing nature of these bacteria in anaerobic culture, *C. acnes* is likely an under-recognized and under-reported etiology of spine infections. Clinicians should be aware of the pathological potential and atypical presentation of *C. acnes* vertebral osteomyelitis.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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

The author(s) confirms 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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How to cite this article: Omosor E, Milosavljevic E, Lawson E, Lopez-Gonzalez MA. Isolated cervical *Cutibacterium acnes* osteomyelitis in a patient with no primary source of infection – A case report and review of the literature. Surg Neurol Int 2023;14:358.