

An Atypical Presentation of Early Periprosthetic Infection After Cervical Disc Arthroplasty

A Case Report

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Abstract

Case: A 22-year-old woman with Klippel-Feil syndrome who underwent cervical disc arthroplasty (CDA) presented 3 months postoperatively with worsening neck pain and radiculopathy. Work-up was negative for infection, but single-photon emission computed tomography revealed increased metabolic activity in the vertebral body below the implant. During revision, the implant was grossly loose and multiple cultures grew *Cutibacterium acnes*. She was treated with an antibiotic course and conversion to anterior fusion without recurrence.

Conclusion: This report highlights the rare presentation of an early periprosthetic infection after CDA caused by *C. acnes*.

Most complications from cervical disc arthroplasty (CDA) can be attributed to the anterior approach or patient selection/surgical technique errors¹⁻³. Deep infection after CDA is rare and usually stems from missed esophageal injury. This report of implant-related infection after CDA is the sixth in the literature, and there is limited under-

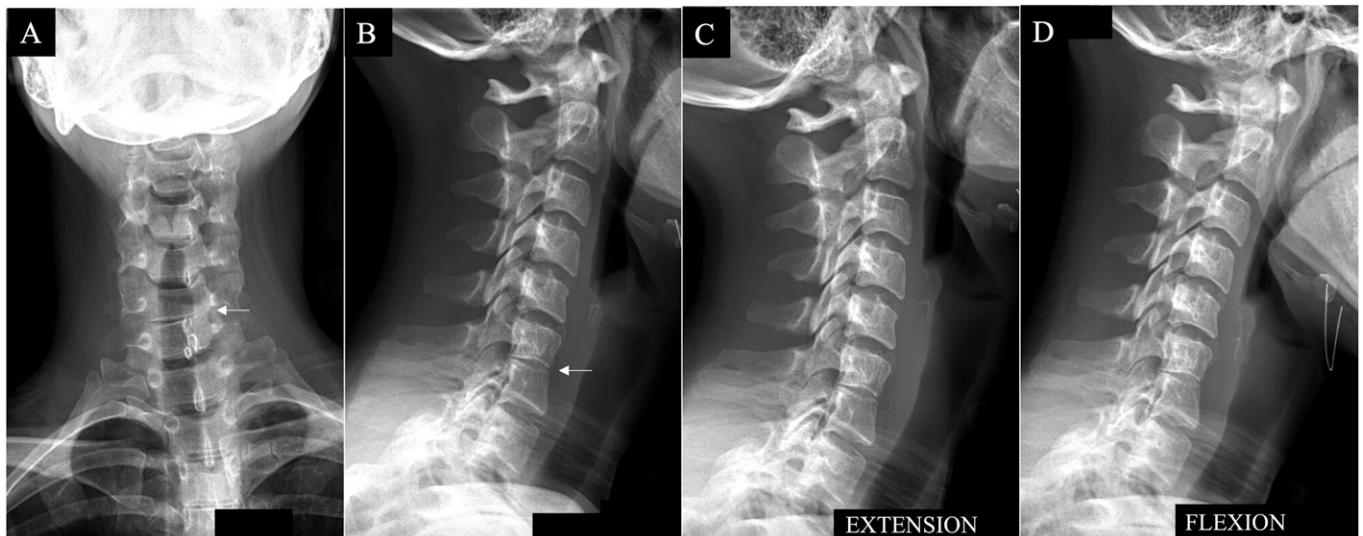


Fig. 1
Preoperative cervical spine radiographs. Anterior-posterior view (**Fig. 1-A**) demonstrating mild rotary dextroscoliosis of the lower cervical spine presumably related to left-sided partial fusion of C6-C7 (arrow). Lateral view (**Fig. 1-B**) showing reversal of cervical lordosis with apex at C5 and wasp-waist sign at C6-C7 (arrow). Extension (**Fig. 1-C**) and flexion (**Fig. 1-D**) views demonstrate mild 2 mm anterolisthesis of C2-C3, C3-C4, and C4-C5 without dynamic instability.

Disclosure: The Disclosure of Potential Conflicts of Interest forms are provided with the online version of the article (<http://links.lww.com/JBJS/C116>).

Keywords young adult; female; infection; *C. acnes*; cervical disc replacement; cervical; artificial disc replacement; loosening; Klippel-Feil syndrome; pain; SPECT-CT; cervical fusion

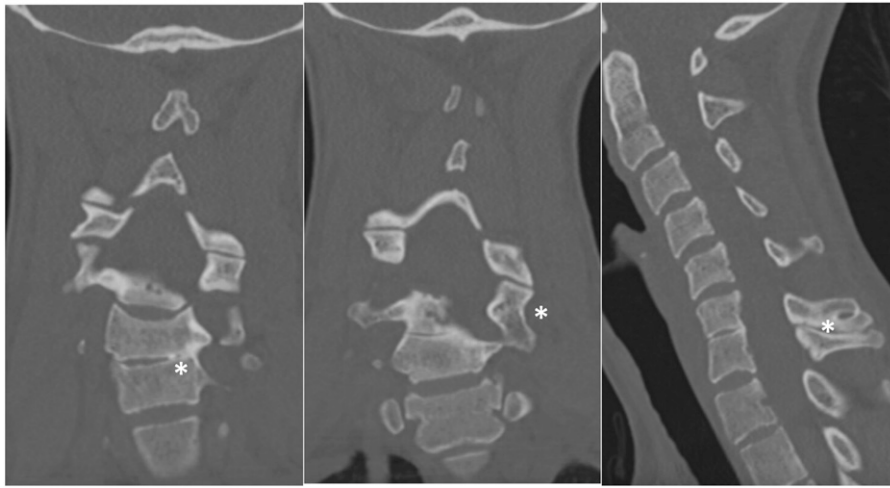


Fig. 2
Preoperative computed tomography coronal and sagittal images of the cervical spine demonstrating hypoplastic C6-C7 disc space with partial ankylosis of the disc space and fusion of the left-sided facets and lamina (*) consistent with Klippel-Feil anomaly.

standing of its presentation, diagnosis, and treatment. We present a case of CDA failure due to infection with an atypical pathogen in a patient with Klippel-Feil syndrome (KFS), along with a review of the literature.

The patient was informed that data concerning the case would be submitted for publication, and she provided consent.

Case Report

A 22-year-old woman with KFS of C6-C7 presented with neck and right arm pain with radiculopathy after a motor vehicle collision. On examination, she had normal strength throughout and decreased sensation in the right C6 nerve root distribution.

Preoperative Imaging

Initial cervical x-rays showed partial fusion of C6-C7 and mild cervical dextroscoliosis and reversal of cervical lordosis without dynamic instability (Fig. 1). A computed tomography (CT)

scan better characterized the segmentation anomaly involving autofusion of the left C6-C7 facet joint and lamina with a hypoplastic and partially ankylosed disc space (Fig. 2). Subsequent magnetic resonance imaging (MRI) revealed multilevel disc degeneration most severe at C5-C6 with annular tear and right foraminal encroachment (Fig. 3). Additional electromyography/nerve conduction study showed electrodiagnostic evidence of acute right C6 radiculopathy.

Clinical Course

The patient had failed conservative treatment including non-steroidal anti-inflammatory drugs, physical therapy, and chiropractic adjustments. Multiple bilateral C5 and C6 medial branch blocks had provided temporary relief but were no longer effective. The patient underwent C5-C6 artificial disc replacement with Prestige LP (Medtronic) and bilateral C5-C6 foraminotomies. The patient's symptoms resolved for

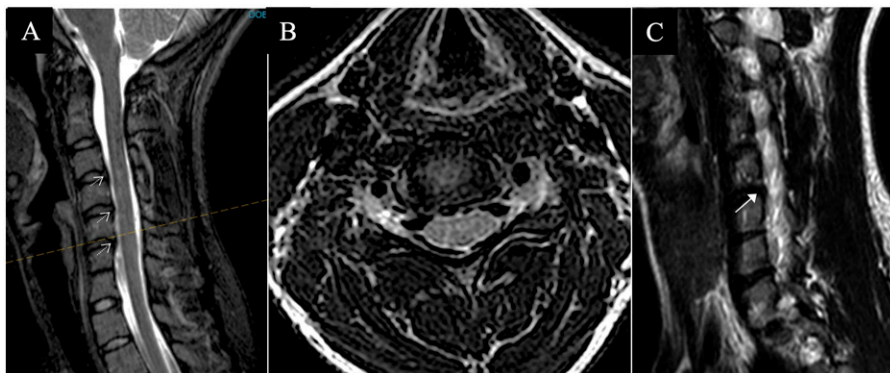


Fig. 3
Preoperative magnetic resonance imaging T2-weighted sagittal and axial images of the cervical spine showing loss of cervical lordosis and segmentation and partial fusion anomaly of C6-C7 vertebrae with rudimentary disc. There are multilevel posterior disc bulges (**Fig. 3-A**: dashed arrows) most severe at C5-C6 with corresponding axial (**Fig. 3-B**) and right sagittal (**Fig. 3-C**) images showing an annular tear and right foraminal encroachment (solid arrow).

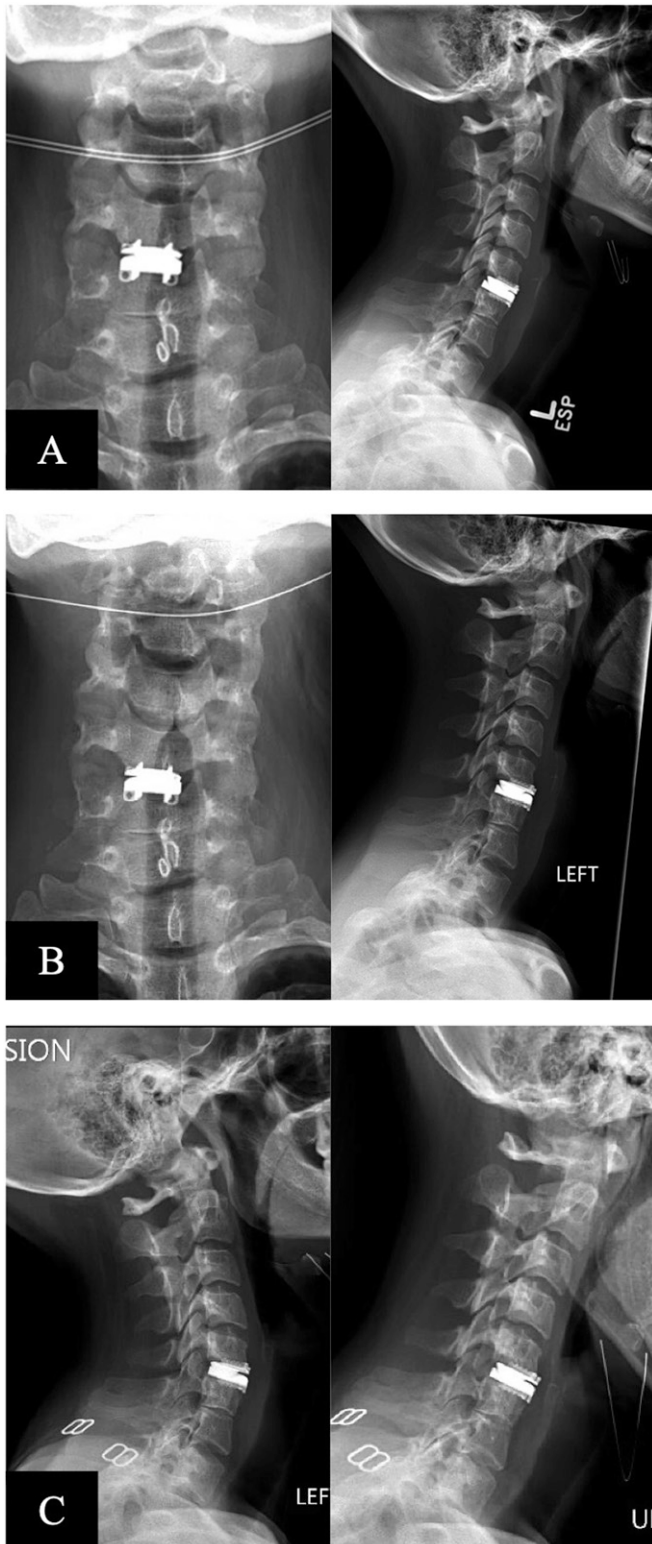


Fig. 4
After C5-C6 cervical disc arthroplasty with prestige LP implant, upright anterior-posterior and lateral cervical spine radiographs were obtained postoperatively at 2 weeks (Fig. 4-A) and 2 months (Fig. 4-B). Flexion/extension views (Fig. 4-C) were obtained at 4 months postoperatively.

approximately 3 months but then returned with a new complaint of left arm C5 radiculopathy. A review of serial postoperative imaging at 4 months showed progressive return of her malalignment without instability (Fig. 4). Repeat CT showed no evidence of obvious infection or implant complication (Fig. 5), but MRI noted progression of C4-C5 disc degeneration with lateral recesses involvement (Fig. 6). Further imaging with single-photon emission computed tomography (SPECT-CT) revealed increased metabolic activity of the C5 and C6 endplates near the implant suggestive of aseptic loosening (Fig. 7). Infection was also considered; however, throughout her clinical course, the patient had no fever, chills, wound complications, or elevated inflammatory markers. Laboratory results showed a white blood count of 7.78 1,000/UL, C-reactive protein of 1.6 mg/L (reference <5.1 mg/L), and sedimentation rate of 9 mm/h (reference <20 mm/h).

Given her intractable pain despite additional facet blocks, the patient elected for C4-C5 CDA and revision C5-C6 CDA. Intraoperatively, the bone endplates appeared and felt normal without any signs of infection, but the lower endplate of the C5-C6 implant was noted to be grossly loose. There was low suspicion for infection and at that time failed osseous integration of the C6 implant surface was highest on our differential diagnosis. Four intraoperative cultures were collected of the endplates and the explanted device. The C5-C6 level was revised to an M6 artificial disc (Orthofix) and primary C4-C5 CDA was performed using a Simplify artificial disc (Nuvasive) (Fig. 8). Intraoperative cultures were held for 3 weeks, and all 4 plates grew single colonies of *Cutibacterium acnes*. The patient was thereafter treated with ceftriaxone 1 g intravenous for 4 weeks followed by 12 weeks of per oral doxycycline. Before culture positivity, the patient had only received surgical antibiotic prophylaxis. No surgical complications were otherwise noted, and the patient followed an expected postoperative course.

Despite completing her course of antibiotics, the patient continued to exhibit progressively worsening neck and bilateral shoulder pain. Repeat SPECT-CT showed persistent increased metabolic activity in the C6 vertebral body and implant endplates of greater intensity than observed preoperatively (Fig. 9). The patient's inflammatory markers remained within normal limits, but given her worsening symptoms and concern for persistent infection, the patient elected to have implants removed and cervical fusion performed at C4-C6. After an uncomplicated anterior cervical discectomy and fusion (ACDF) (Fig. 10), the patient continued through an expected postoperative course with ultimate resolution of symptoms.

Discussion

The popularity of CDA as a motion-preserving treatment of degenerative disc disease has increased since its introduction in the early 2000s, with favorable outcomes compared with ACDF⁴⁻¹⁰. Given that it is a relatively new technology, our understanding of its durability and potential complications that may arise in the future is limited.

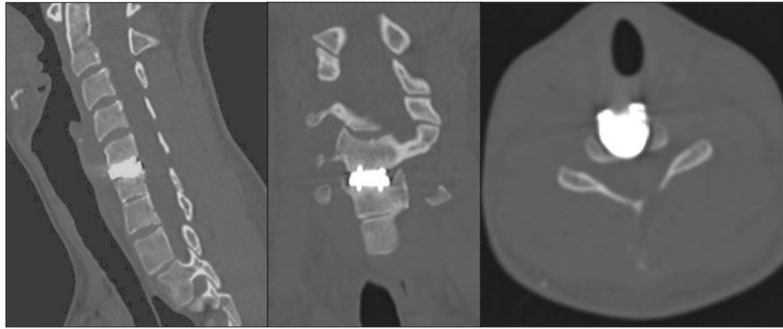


Fig. 5

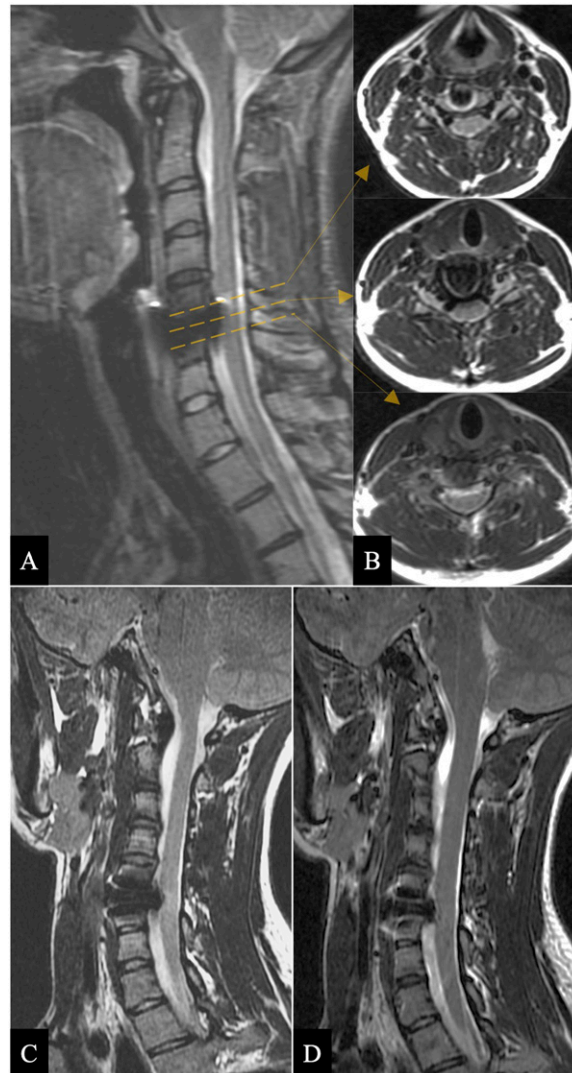


Fig. 6

Fig. 5 Cervical computed tomography without intravenous contrast images obtained 4 months postoperatively showing C5-C6 implant appropriately positioned with good integration of bone-implant interface. Subtle lucency along the anterior superior endplate of C6 is visualized only on the sagittal view. No radiographic evidence of adjacent soft-tissue mass or fluid collection, osteolysis, or implant loosening. **Fig. 6** Postoperative cervical spine magnetic resonance imaging including (**Fig. 6-A**) sagittal short-T1 inversion recovery sequence with corresponding (**Fig. 6-B**) metal artifact reduction sequence axial images show normal bone marrow signal and no evidence of soft-tissue mass or fluid collection adjacent to C5-C6 CDA. Metallic artifact obscures the visualization of the central canal and neural foramen at the C5-C6 level. There is progressive disc degeneration of the adjacent C4-C5 level involving lateral recess disc herniations as seen on the T2-weighted left sagittal oblique (**Fig. 6-C**) and right sagittal oblique (**Fig. 6-D**) views. CDA = cervical disc arthroplasty.

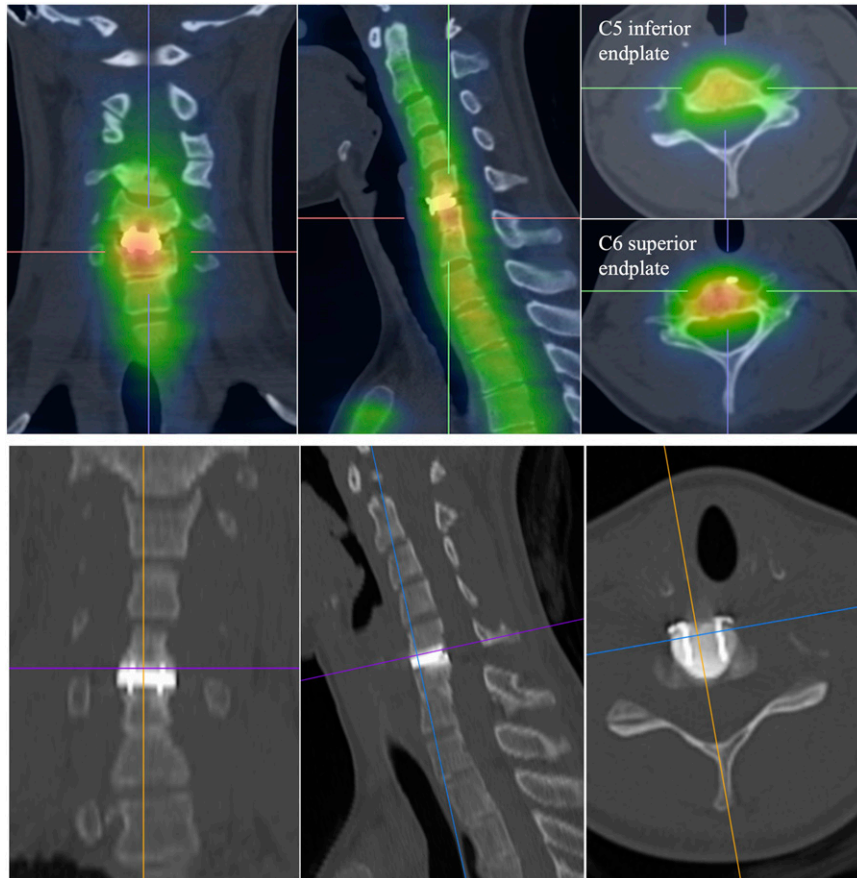


Fig. 7



Fig. 8

Fig. 7 Cervical CT with SPECT obtained 6 months after index surgery. **Top** 3-dimensional reconstruction images demonstrating increased metabolic activity at the C5-C6 implant endplates with the highest activity localized at the superior endplate of C6. **Bottom** CT-only portion showing lucency of the bone-surface interface between the implant's inferior endplate and the superior endplate of C6. Taken together, these findings suggest implant loosening of the inferior endplate of the implant. SPECT-CT = single-photon emission computed tomography. **Fig. 8** Postoperative cervical spine radiographs 3 months after revision C5-C6 CDA (M6 artificial disc) and primary C4-C5 CDA (Simplify artificial disc). Radiographs show a similar appearance of reversal of the cervical lordosis and appropriate hardware position with no evidence of loosening. Dynamic radiographs show limited yet preserved motion at the operative levels. CDA = cervical disc arthroplasty.

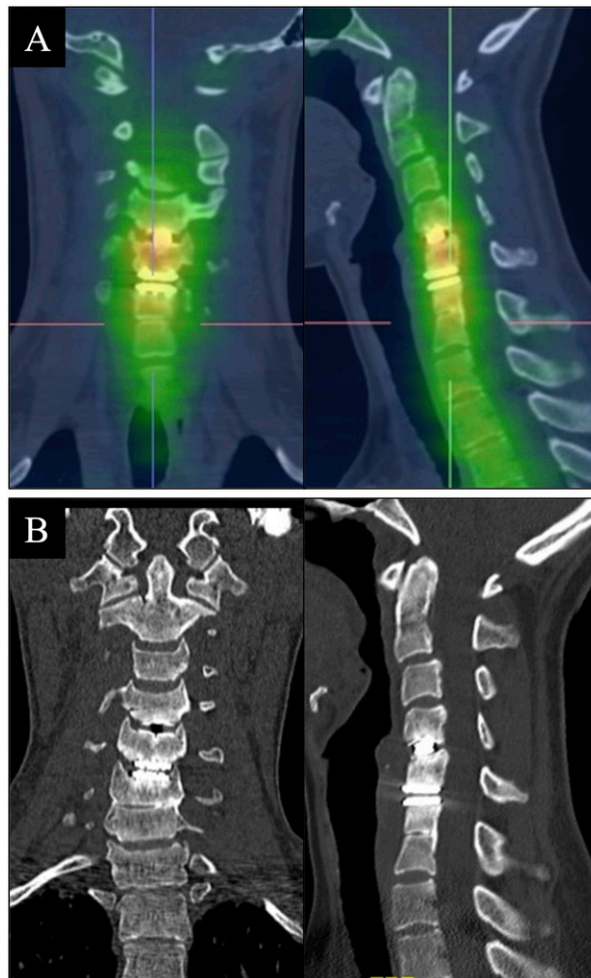


Fig. 9



Fig. 10

Fig. 9 Repeat SPECT-CT obtained 4 months after revision. **Fig. 9-A** Coronal and sagittal SPECT views demonstrating persistently increased metabolic activity of the C6 superior endplate as well as new, higher signal intensity within the C5 vertebral body and adjacent to the C4-C5 artificial disc implant. **Fig. 9-B** The same views on plain CT indicate likely subsidence of the C4-C5 implant with intrusion into the C4 inferior endplate and resultant mild kyphotic deformity with an apex at C4. The prevertebral soft tissues seem normal with no evidence of abnormal metabolic activity, mass, or abscess. SPECT-CT = single-photon emission computed tomography. **Fig. 10** Postoperative radiographs 8 months status after C4-C6 implant removal and conversion to anterior cervical fusion demonstrating restoration of cervical lordosis and stable hardware with interval bony fusion of C4-C5 and C5-C6.

Short-term complications of CDA are commonly related to the anterior cervical approach, similar to those seen in ACDF^{1,11}. In the mid-term to long-term period, CDA may have unique complications such as heterotopic ossification and osteolysis, but most often patients are asymptomatic and do not require treatment^{3,12-14}. However, unlike other joint replacement procedures, the risk of delayed implant-related infection after CDA is extremely low¹⁵⁻¹⁷. An analysis of 7,627 patients undergoing cervical spine surgery reported no deep infections after

CDA¹⁷. Hisey et al. reported 18.4% rate of wound infections in 179 patients with the Mobi-C cervical disc (Zimmer Biomet); however, none of them were deep infections⁵. Anderson et al.¹⁴ reported that of 242 patients who underwent Bryan disc arthroplasty (Medtronic Sofamor Danek), 7 (2.8%) had superficial skin infections treated successfully with oral antibiotics, and no patients had deep infections. Cuellar et al. reported only 1 superficial infection of 147 (0.7%) outpatient CDAs¹⁸. A meta-analysis¹⁵ and systematic review¹¹ found that infection-related

TABLE 1 Case Summaries^{19,20*}

	Case 1	Case 2	Case 3	Case 4	Case 5
Age (yrs)/gender	59/male	63/male	68/male	51/male	48/male
Previous procedure	C6-7 CDA	C4-5 CDA	C4-5 ACDF, C5-6 CDA	C5-6, C6-7 CDA	C5-6, C6-7 CDA
Time to presentation (yrs)	5	4	7	5	2
Symptoms	Progressive dysphagia, palpable neck mass	Progressive dysphagia	Rapidly progressive dysphagia	Dysphagia and worsening respiratory compromise	Dysphagia, neck swelling, and draining sinus
Inflammatory markers	Normal	Normal	Normal	Normal	Normal
CT/MRI cervical spine findings	Contrast enhancing prevertebral collection anterior to implant	Enhancing soft-tissue mass with fluid collection anterior to implant	Prevertebral fluid collection contiguous with C5-6 implant; osteolysis of vertebral body endplates	Retropharyngeal collection anterior to implant with compression of esophagus and surrounding structures	Superficial fluid collection tracking deep to C5-C7 with osteomyelitis; implant endplate lucencies
Operative findings	Necrotic debris; implant failure with disintegration of artificial sheath and annulus	Purulent abscess surrounding implant; no implant loosening	Encapsulated soft-tissue collection	Purulent fluid collection surrounding implant	—
Surgical procedure	Implant removal, I&D, C6-7 fusion using cage, and bone allograft	Implant removal, I&D, and immobilization in cervical collar for 6 wk (no fusion)	Implant removal, fusion of C4-6 with autologous bone graft and anterior plating	Wound exploration, I&D. Prosthesis left in situ	Wound exploration, I&D. Prosthesis left in situ
Microbiology	<i>P. acnes</i> growth on culture	<i>P. acnes</i> isolated by 16S rRNA molecular sequencing of disc space	Cultures no growth. No 16S rRNA molecular sequencing performed	Culture no growth. <i>P. acnes</i> isolated on 16S rRNA molecular sequencing	<i>P. acnes</i> isolated on tissue culture and 16S rRNA molecular sequencing
Post-operative course and outcome	6 wk IV benzylpenicillin and moxifloxacin \bar{r} suppressive PO amoxicillin. Dysphagia resolved, no recurrence, or additional intervention	5 d IV teicoplanin \bar{r} 6 wk IV amoxicillin and rifampicin. Dysphagia resolved, no recurrence, or additional intervention	48 h of prophylactic cefuroxime. Dysphagia resolved, no recurrence, or additional intervention	1 wk IV amoxicillin/linezolid \bar{r} 6 wk IV ceftriaxone/PO linezolid \bar{r} 1 yr PO doxycycline and rifampicin. Residual mass asymptomatic and no additional intervention	6 mo of multidrug antibiotic regimen \bar{r} 6 mo clindamycin PO. Required hyperbaric oxygen therapy for wound drainage. Asymptomatic after 32 mo

*ACDF = anterior cervical discectomy and fusion, CDA = cervical disc arthroplasty, CT = computed tomography, I&D = irrigation and debridement, IV = intravenous, MRI = magnetic resonance imaging, PO = per oral, and rRNA = ribosomal RNA.

complications after CDA ranged between 1.2% and 22.5%; however, most reports were superficial infections, and no study reported any implant infection requiring surgical intervention within 2 years of surgery.

This is the sixth reported case of infection-related failure of CDA (Table I). The first case was reported by Xia and Winder in 2019, followed by a case series of 4 patients presented by Harris et al.^{19,20}. All prior reports presented with late infection 2 to 7 years postoperatively with dysphagia, a prevertebral mass, and normal inflammatory markers. These infections all involved the M6-C cervical disc replacement and, in addition to intravenous antibiotics, were treated with implant removal and fusion in 3 cases and irrigation and debridement with implant retention in the others. *Propionibacterium acnes* (now *C. acnes*) was identified in 4 cases. Literature suggests that *C. acnes* susceptibility to commonly prescribed antibiotics—beta-lactams, quinolones, rifampicin, and clindamycin—is quite favorable, with recommended treatment for prosthetic joint infection being 3 months of bitherapy—a 2-week to 6-week intravenous phase followed by orals for the remainder of the course^{21,22}.

Our case differs from previous reports in that the patient presented early with symptoms only a few months postoperatively, with no accompanying soft-tissue mass or signs of infection. Despite no clear indications in their medical history or initial presentation, increased endplate activity seen on SPECT-CT raised the possibility of an underlying infection. Consistent with the low virulence associated with *C. acnes*, no purulence or obvious infection was encountered intraoperatively; however, 1 of the endplates had loosened or failed to integrate.

The current case poses several questions that warrant further discussion, such as the optimal treatment for neck pain and cervical radiculopathy in a patient with KFS. Preserving motion adjacent to a congenital fusion is believed to be advantageous over ACDF by limiting the degeneration of subsequent levels and supported by several reports²³⁻²⁷. However, patient selection remains a crucial factor in all CDA procedures, and alternative motion-sparing options such as posterior cervical laminoforaminotomy should also be considered when appropriate²⁸.

In evaluating for osteomyelitis, fluid-sensitive MRI sequences are the most sensitive, but metal artifacts from implant materials can hinder accurate assessment of the surrounding

bones and neurological structures, as illustrated in Figure 6. Hence, the imaging characteristics of implants are an important factor to consider in decision-making, the most favorable being titanium-coated polyetheretherketone-on-ceramic implants currently, which produce virtually no artifact on MRI^{29,30}. In cases where MRI results are inconclusive, SPECT/CT scans have proven to be a reliable diagnostic tool for identifying pseudoarthrosis, implant loosening, adjacent segment degeneration, osteomyelitis, and device-related infections³¹⁻³⁴. Its application in these areas will likely expand as advancements in nuclear imaging technology and research continue to develop.

The identification of *C. acnes* as the cause of infection is a clear similarity to previously reported cases. *C. acnes* has emerged as a key player in spine pathology having been implicated in native and implant-associated infections and suspected agent in the Modic changes seen in disc degeneration. *C. acnes* is most associated with latent postoperative spine infections with corresponding soft-tissue reactions. Our case suggests *C. acnes* may also lead to early implant loosening without other signs of infection. Given the occult presentation and the difficulties associated with isolating *C. acnes*, this complication of CDA may be underreported. Thus, in cases of CDA complaints of unknown origin, there should be a low threshold for intraoperative culture for *C. acnes* during revision CDA. ■

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