Iatrogenic pyogenic osteomyelitis of C-1 and C-2 treated with transoral decompression and delayed posterior occipitocervical arthrodesis

Case report

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The authors describe a case of osteomyelitis of the craniovertebral junction caused by iatrogenic infection of the spine during corticosteroid injection therapy. This 58-year-old diabetic man presented with acute exacerbation of neck pain that had begun 4 months prior to admission. He did not experience the associated fever, chills, or sweats, but he did notice transient weakness in the right upper extremity. A computed tomography (CT) scan of the cervical spine demonstrated a destructive process involving the odontoid and the left occipitocervical and atlantoaxial joints that was not present on a CT obtained 2 months earlier, just before trigger-point and left-sided C1–2 facet joint corticosteroid injections. A diagnosis of staphylococcal osteomyelitis was made, and initial treatment with external immobilization and appropriate antibiotic therapy failed to control radiographically demonstrated and clinical progression. The patient was successfully treated using staged anterior decompression and posterior instrumented fusion with prolonged antibiotic therapy.

To the authors' knowledge this case is the first reported instance of iatrogenic pyogenic osteomyelitis of the craniovertebral junction successfully treated with anterior decompression and delayed posterior arthrodesis.

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KEY WORDS • arthrodesis • cervical spine • infection • pyogenic osteomyelitis
left C1–2 facet joint injection were administered. These injections were unsuccessful in relieving his symptoms, and the patient declined further injections. He was also treated with a short course of oral steroids for possible temporal arteritis, but these agents were discontinued after a negative biopsy. Despite the addition of Lidoderm patches, extended-release and short-acting oral narcotics, gabapentin, and muscle relaxants, the pain acutely worsened, necessitating inpatient admission.

Physical examination revealed an afebrile obese patient; there was tenderness to palpation of the upper cervical spine and an associated decreased range of movement. There was no weakness to confrontational muscle testing, no sensory deficits, and no upper motor neuron signs. Results of laboratory studies revealed a white blood cell count of 14.5 × 10^9/L, a CRP of 115 mg/L, and an ESR of 109 mm/hour. Purified protein derivative testing was negative. A CT scan of the cervical spine demonstrated lytic destruction of the left atlantooccipital joint, atlantoaxial joint, and the tip of the odontoid, clearly none of which was present on the scan obtained 2 months earlier (Fig. 1).

Computed tomography scans of the chest, abdomen, and pelvis did not reveal a primary malignancy. The patient was initially placed in a Miami J collar, and an empirical antibiotic regimen of vancomycin and ciprofloxacin was administered. Computed tomography-guided aspiration was considered for both therapeutic and diagnostic purposes; however, we decided that it was not necessary as the patient was stable, and blood cultures drawn in the emergency department subsequently revealed methicillin-sensitive *Staphylococcus aureus*. Therefore, nonsurgical management was continued with nafcillin. An MR image obtained after a 1-week antibiotic regimen revealed an epidural collection dorsal to the odontoid as well as soft-tissue swelling involving the prevertebral space extending from the skull base to C4–5. After 3 weeks of antibiotics and external immobilization, the patient reported progressive neck pain and transient numbness and weakness in his upper extremities while sitting. Repeated serologic tests revealed a white blood cell count of 11.1 × 10^9/L, CRP of 142 mg/L, and ESR of 126 mm/hour. Comparison MR images of the cervical spine demonstrated an increase in the epidural phlegmon with abscess formation and increased T2 signal intensity at the cervicomedullary junction (Fig. 2C). A CT-guided aspiration was considered; however, the neuroradiologists believed the liquid component was so minor that its removal would not lead to satisfactory decompression. Thus, in view of the patient’s clinical symptoms, worsening inflammatory parameters, and radiographically demonstrated progression, surgical intervention was recommended.

**First Operation.** The patient underwent a planned two-stage procedure. Stage 1 included a tracheostomy followed by a transoral transpalatal approach with odontoidectomy and debridement of the epidural abscess to decompress the spinal cord and brainstem, followed by halo vest application. After removing the anterior ring of C-1, the base of the odontoid was transected using a high-speed diamond bur allowing mobilization of the mottled tip of C-2 away from
the cervicomedullary junction. The dense inflammatory reaction made adequate decompression impossible without partial durotomy and cerebrospinal fluid leakage, which was repaired with the application of Duragen and fibrin glue. A lumbar drain was successfully placed at the conclusion of the procedure.

First Postoperative Course. Specimens were sent for both microbiological and histopathological analysis. Gram staining and cultures were nondiagnostic, including those for tuberculosis and fungal infections. Histopathological analysis revealed osteomyelitis of the odontoid with both acute and chronic inflammatory components. A postoperative CT scan showed satisfactory anterior decompression. The lumbar drain was used to decompress the dural repair and was removed without incident after 10 days. The patient advanced slowly to a regular diet over the next week and had minimal dysphagia.

Second Operation. Twenty-five days after the first operation the patient underwent Stage 2 of the procedure, a posterior occipitocervical arthrodesis (occiput to C-4) with instrumentation (SUMMIT, DePuy AcroMed, Inc.) and a combination of iliac crest autograft, allograft, and recombinant bone morphogenetic protein–2 (INFUSE, Medtronic Sofamor Danek). The halo vest was removed, and the patient was placed in a Miami J collar.

Second Postoperative Course. The patient had no focal neurological symptoms after surgery. The preoperative axial neck pain rapidly improved. The tracheostomy tube was removed, and the patient was discharged to a rehabilitation facility 6 days after the posterior operation and went home 2 weeks thereafter. The orthosis was removed after a 3-month follow-up, and moxifloxacin and rifampin were orally administered for a total of 6 months, with normalization of inflammatory parameters. Follow-up plain radiographs (Fig. 3) and a thin-cut CT demonstrated stable positioning of the instrumentation and a solid sheet of bone from the left atlantooccipital and atlantoaxial joints on the left extending to C-4.

Discussion

There has been a recent increase in the use of corticosteroid injection therapy in the multidisciplinary management of painful degenerative spinal conditions in the cervical, thoracic, and lumbar regions. This therapy includes medial branch blocks and facet, perifacet, sacroiliac, and epidural steroid injections. Infectious complications have been sporadically reported in the literature and, in fact, may be underreported. Infection associated with epidural anesthesia, particularly in conjunction with indwelling catheters, has been more thoroughly documented but is still believed to be relatively rare. A retrospective review of over 4000 spinal anesthetics performed at the Mayo Clinic revealed only two infectious complications. In 2006 Hooten et al performed a MEDLINE literature review to determine the incidence of infectious complications in interventional spinal procedures. The study period covered from 1966 to 2004, and the results revealed only 21 infectious complications after epidural injections, four after facet injections, and one associated with a sacroiliac joint injection.

Fig. 2. Sagittal T1-weighted MR images obtained on initial presentation (A) and 12 weeks later (B), demonstrating progression of epidural phlegmon with abscess formation despite 3 weeks of antibiotic treatment. Sagittal T2-weighted MR image (C) revealing increased mass effect and hyperintensity at the cervicomedullary junction as well as the pre-vertebral hyperintensity down to C-5.
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These authors concurrently described the first reported instance of lumbar discitis after epidural corticosteroid injection. Falagas and colleagues subsequently described the first case of lumbar discitis after facet joint injection and Weingarten et al. reported the first case of septic lumbar facet arthritis after injection at an adjacent joint. Only Waldman and Huang et al. have reported cervical epidural abscess after epidural steroid injections; instances described by both authors involved the subaxial spine. Elias reported a single case of subaxial epidural abscess after a trigger point injection. Halla and associates and Sasaki et al. have both reported septic arthritis of the atlantoaxial joint, but these cases did not involve prior injection therapy. Yazawa and colleagues described a case of cervical epidural abscess and osteomyelitis of C-1 and C-2 after acupuncture treatment for chronic neck pain. To our knowledge, the case in the present report is the first instance of iatrogenic osteomyelitis with associated epidural abscess at the craniocervical junction due to intraarticular corticosteroid injection.

Making a clinical diagnosis of an SEA with or without associated osteomyelitis can be difficult in the setting of acute and chronic neck pain. The clinical triad of back or neck pain, fever, and neurological deficit may not always be present and depends on the stage of the infection. The clinical course has been classified into four stages: spinal ache, root pain, weakness, and paralysis. In patients who present for interventional spinal procedures, this diagnostic dilemma is amplified because many present with Stage 1 and 2 symptoms at baseline. The lack of a classic radicular pain pattern at the craniocervical junction is also problematic. The patient in the current case presented with severe symptoms approximately 2 months after his procedure, but this time course can vary from days to months. This variation underscores the importance of a high index of suspicion in patients with predisposing conditions (diabetes mellitus, alcoholism, or immunodeficiency), particularly those with escalating postinjection pain. In a retrospective review by Tang et al. of 46 patients with SEA, nearly half were diabetic.

The clinical diagnosis in the current case was confirmed with a combination of laboratory and imaging studies. Serological markers of inflammation (ESR and CRP) are almost uniformly elevated, and peripheral leukocytosis is seen in approximately two thirds of patients with SEAs in large studies. Bacteremia is associated with more than half of patients with SEA, particularly in cases due to Staphylococcus aureus.

Gadolinium-enhanced MR imaging is widely considered as the gold-standard imaging modality. In the present case the extent of infection was readily identifiable on MR images. The decision to proceed with aggressive surgical intervention was greatly facilitated by the dramatic change in anterior compression and the T2-weighted signal change on the follow-up study (Fig. 2), which would have been much less readily apparent using other modalities.

The necessity of surgical intervention for SEA is somewhat controversial. Open surgical drainage with subsequent long-term intravenous antibiotics is the generally favored treatment, although successful medical management has been reported. An initial course of intravenous antibiotics can be considered if patients are neurologically stable. Identification of the offending organism can be made via blood cultures or needle biopsy sampling if patients are too unstable or frail for surgery. For patients with significant or progressive neurological deficits, the timing of surgical intervention is critical. Results of a retrospective review of patients with SEA by Darouiche et al. showed neurological improvement in 85.7% of paralyzed patients who had undergone surgery within 24 hours of onset compared with 14.3% of those surgically treated more than 72 hours after paralytic onset. Based on a retrospective review of 41 cases of SEA of various origins, Khanna et al. proposed a grading scale to predict outcome. An age younger than 60 years, less than 50% compression of the thecal sac, and a shorter than 72-hour duration of neurological symptoms were cutoff variables used to assign patients a grade of 0 to III. A significant increase in the percentage of patients with a poor clinical outcome was seen between Grades I (33%)
and II (72.7%). Given the significant biomechanical consequences of occipitocervical fusion, a positive bacteriological diagnosis, and a paucity of neurological symptoms at the time of presentation, we believed that a trial of medical management and close observation were reasonable in this case. Although the patient had no significant objective change on neurological examination, progression of both pain and radiographically demonstrated thecal sac compression by an enlarging abscess and worsening of laboratory parameters of inflammation mandated prompt surgical intervention.

Conclusions

Pyogenic osteomyelitis and epidural abscess involving the craniocervical junction constitute an extremely uncommon complication of interventional spinal procedures. Diagnosis requires a high index of suspicion and timely medical and surgical intervention. Patients with risk factors for infectious complications, particularly those with diabetes mellitus, should be monitored closely for new or worsening symptoms and should be well informed of the potential risks.

Disclaimer

Neither of the authors has a financial interest in the subject under discussion.

References


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