CASE REPORT

Severe rotational deformity, quadriparesis and respiratory embarrassment due to osteomyelitis at the occipito-atlantoaxial junction

We present the case of an 83-year-old man who developed quadriparesis and respiratory embarrassment following osteomyelitis at the occipito-atlantoaxial junction. He had developed an abscess at this site after an earlier urinary infection with methicillin-resistant staphylococcus aureus. Stabilisation of the neck and antibiotic therapy led to an almost complete neurological recovery without recourse to anterior surgery.

The incidence of pyogenic spinal osteomyelitis has increased with the rising elderly population, immuno-compromised hosts and intravenous drug abusers. Malawski and Lukawski reviewed 442 patients with pyogenic spinal spondylitis and noted that the cervical spine was affected in only 5.8% and the C1 or C2 levels were affected in only 0.7%. Before 1990, approximately one-third of patients with osteomyelitis of the odontoid process died. This was probably because early diagnosis was difficult before the introduction of MRI. Thereafter, the prognosis for osteomyelitis at this site improved, in part due to earlier diagnosis but also because the introduction of good implants facilitated occiput-cervical arthrodesis.

The anatomy of the craniocervical junction is more complex than subaxial regions. Halla et al reported torticollis in association with pyogenic arthritis of the C1-2 lateral facet joint. We are not aware of any report of osteomyelitis between the occiput and axis causing severe rotational deformity, quadriparesis and respiratory embarrassment. We report such a case in an elderly man following methicillin-resistant Staphylococcus aureus (MRSA) infection in the urinary tract, and discuss the management of this condition.

Case report

An 83-year-old man with an indwelling urinary catheter for the management of urinary retention caused by prostatomegaly, was admitted to a local hospital because of fever. Since MRSA was detected on urine culture, vancomycin was administered intravenously at a dose of 0.5 g twice a day for 12 days. He complained of mild pain in the cervical region. The urinary tract infection was eliminated after one month. However, nine days after discharge he developed quadriparesis. The neurologist in the admitting hospital suspected a cerebrovascular accident, because the quadriparesis predominantly affected the right arm and the patient already had a mild hemiplegia on the left side after a previous cerebral infarct five months earlier. When the radiologist helped the patient to arise from the scanning table after cranial CT, he stopped breathing. He was resuscitated and respiration returned. The cervical spine was noted to be rotationally deformed to the right and an attempt to reposition the neck to a neutral position caused dyspnoea. Therefore, he remained with his neck severely rotated. As the CRP was 12.9 mg/dl, intravenous vancomycin was recommended at a dose of 0.5 g twice a day for 12 days. Although his general condition gradually improved the severe rotational deformity persisted. Therefore, he was referred to our hospital two weeks after the onset of quadriparesis.

On admission he had a temperature of 37.6°C, percutaneous oxygen saturation of 95% and severe torticollis with his head rotated to the right by approximately 70°. He had marked weakness in his arms and a less marked weakness in his legs. There was bilateral sensory disturbance below the C3 level.

CT demonstrated severe bone destruction of the occipito-atlantoaxial complex and marked rotation at the atlantoaxial junction (Fig. 1). MRI demonstrated a heterogeneous lesion on T1- and T2-weighted images in the region between the atlantoaxial joint and retropharyngeal space.
A halo ring was applied and manual reduction of the atlantoaxial rotation attempted with the patient conscious. The neck was slowly reduced toward a neutral position while monitoring the percutaneous oxygen saturation. Reduction was achieved without dyspnoea or deterioration in his neurological status. A halo-vest was then applied to maintain the alignment. Resection of the posterior arch of the atlas and posterior arthodesis between the occiput and C4 was performed. Pedicular screws were inserted at C3 and C4 using the Olerud Cervical System (Anatomica, Gothenburg, Sweden), but not into the atlas or axis, which were infected. Anterior surgery was not undertaken because pre-operative MRI showed that the abscess had almost resolved (Fig. 2).

A Philadelphia collar (Össur, Reykjavik, Iceland) was retained for three months post-operatively. Three weeks post-operatively, the level of CRP was 0.35 mg/dl and by six weeks muscle power had recovered considerably and was MRC grade 4 in all limbs. Six months after surgery, he could walk with a frame and at one year, MRI showed complete resolution of the abscess and no compression of the spinal cord. Plain radiographs at three years post-operatively showed complete fusion from the occiput to C4 without disruption of the fixation (Fig. 3). Further improvement continued and at five years he could walk with a stick and undertake activities of daily living independently.

Discussion
Osteomyelitis of the odontoid process had been considered to be rare; however, there have recently been an increasing number of reported cases which can be attributed in part to wider use of MR imaging.\textsuperscript{8-13} Earlier diagnosis with prompt treatment has resulted in improved recovery. Whereas almost all patients with osteomyelitis of the dens reported since 1990 survived and recovered,\textsuperscript{8-13} approximately one-third of those reported before 1990 died.\textsuperscript{4-7} In our patient osteomyelitis extended from the occiput to the axis, including the dens. The craniovertebral junction consists of a set of synovial articulations at the atlanto-occipital, atlantoaxial, and atlanto-odontoid joints, all of which were infected. There is a previous paper presenting five patients with osteomyelitis of the occiput, atlas, and axis\textsuperscript{14} but only one was described as having a rotational deformity. However, the severity was not recorded.\textsuperscript{14} Halla et al\textsuperscript{8} also reported a patient with septic arthritis of the C1-2 lateral facet joint and torticollis. Both these patients with rotational deformity had the sudden onset of
motor weakness. The first developed quadriparesis approximately one month after onset of the rotational deformity. The second developed right-sided hemiparesis over two months after the onset.

In patients in whom infection cannot be controlled or in those with neurological symptoms, curettage of the lesions or decompression should be performed, and simultaneous arthrodesis is often required. Posterior atlantoaxial arthrodesis with instrumentation of the infected vertebral body or facet is unsuitable. Also, if wiring or hook fixation to unaffected vertebral arches or spines is performed in such patients, the fixation remains compromised. However, a pedicle screw supplies sufficient fixation albeit with a risk of injury to the vertebral artery and spinal cord. Several authors performed curettage transorally for patients with odontoid osteomyelitis but this was not required in our case as MRI showed that the abscess had almost resolved when stabilisation was undertaken. Therefore, posterior arthrodesis was sufficient. Zigler et al also reported a good outcome for two of their five patients with isolated posterior arthrodesis and we consider this to be a satisfactory option in the presence of severe destruction of the craniocervical junction, assuming that the anterior abscess is small.

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**References**


