

# C1 and C2 vertebrae osteomyelitis: A misleading presentation leading to a fatal outcome.

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## ABSTRACT

*Cervical vertebral osteomyelitis is rare. While an early and correct diagnosis is critical to prevent catastrophic neurological injury, the diagnosis of cervical vertebral osteomyelitis is often difficult because of its rarity and variable symptoms. We present a case of C1 and C2 vertebrae osteomyelitis with a misleading presentation and its fatal outcome.*

**KEYWORDS:** Cervical spine, osteomyelitis, spinal infection

## INTRODUCTION

Vertebral osteomyelitis is rare and represents only one percent of all cases of bone infection. Of these only three to six percent involve the cervical vertebrae. In the cervical region, C5 and C6 vertebrae are the commonest vertebrae involved which accounted for about 40 percent of cases and isolated cases of the upper cervical are the least common. The diagnosis of cervical vertebral osteomyelitis is often delayed because of its atypical signs and symptoms. Early diagnosis remains a major challenge as it is critical to prevent catastrophic neurological injury and essential for an optimal outcome.

## CASE REPORT

A 72-year-old lady with background history of hypertension and chronic otitis externa of the left ear presented with a two-day history of progressive left-sided weakness of the limbs. She felt unwell and had poor appetite for a week. There was no associated history of fever, neck stiffness, vomiting or fainting episode. Examination revealed a conscious elderly woman with a blood pressure of 142/74 mmHg and Glasgow Coma Score of 15. She has facial asymmetry and right sided deviation of the tongue. The left upper and lower limbs were graded to have 0/5 and 1/5 motor powers respectively. The right upper and lower limbs were neurologically normal. A clinical diagnosis of cerebrovascular accident was made. She was mildly anemic with a hemoglobin level of 9.6 g/dL. Other blood parameters including white blood cell count, platelet count, renal and liver function tests were essentially normal. An urgent plain CT brain was reported as normal.

A day after admission she complained of earache and neck pain with inability to turn her head. She also had dysphagia and difficulty to open her mouth. Two days later she had spikes of fever. Examination revealed

unchanged neurological status but she has bilateral basal lung crepitations. Ear examination was normal. She was treated with empirical antibiotics for a presumptive diagnosis of aspiration pneumonia.

Plain radiograph of the cervical spine was requested for persistent neck pain. It revealed C1-C2 subluxation and osteopenic looking C1-C2 vertebrae. On further enquiry she gave a history of a fall a few years ago and admitted that the facial asymmetry has been existed for several years. She has been bothered with neck pain for the past two months. CT scans of the cervical spine confirmed a C1-C2 subluxation with total absence of the dens, and partial lytic destruction of C1 and C2 vertebral bodies. MRI images of the cervical spine revealed the affected bone and intervertebral disc space to be hypointense on T1-weighted image, hyperintense on T2-weighted image and enhanced post contrast. Additional information of retropharyngeal and epidural collections with anterior sub-ligamentous and paravertebral spread were noted [Figure 1, 2 and 3].

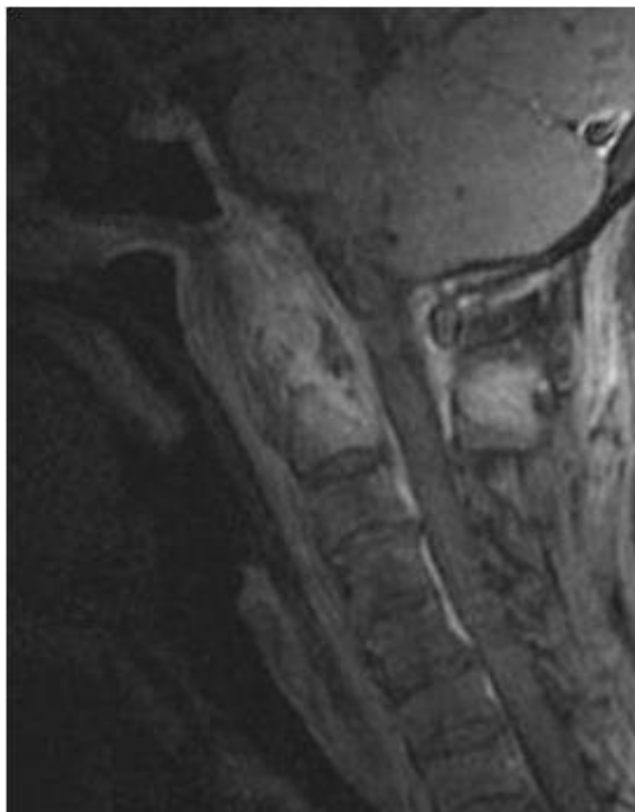
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**Figure 1:** Sagittal T1-Weighted MR image of cervical spine showed hypointense C1 and C2 vertebrae lesion.



**Figure 2:** Sagittal T2-Weighted MR image of cervical spine showed the same lesion to be heterogeneously hyperintense.



**Figure 3:** Post gadolinium image showed enhancement of this C1 and C2 vertebrae lesion. Anterior subligamentous, epidural spread down to C6 level and paravertebral involvement also noted.

She subsequently had an episode of cardiorespiratory arrest due to acute high cord compression syndrome complicated with diaphragmatic paralysis and aspiration pneumonia. Following successful resuscitation she was then transferred to ICU for assisted ventilation and intensive care. Following optimization she underwent C1-C2 decompression and debridement through an anterior approach. Her post operative progress was complicated with persistent sepsis and she eventually succumbed due to septicaemia and renal impairment three weeks later.

Histopathological examination of debrided tissues showed necrotic bone fragments embedded in soft tissue, cartilage and skeletal muscle. There are mixed acute and chronic inflammatory cellular infiltrates without features of epithelioid granuloma or malignant cells. Specimen for culture and sensitivity grew *Actinobacter* sp. and staining for acid fast bacilli was negative.

#### DISCUSSION

Vertebral osteomyelitis is rare and represents only one percent of all cases of bone infections. Of these only three to six percent involve the cervical vertebrae.<sup>1</sup> The diagnosis of cervical vertebrae osteomyelitis is often difficult because of its rarity and variable symptoms. Delayed in diagnosis is a recognized problem of this condition and this can subsequently impose catastrophic implications to the patient.

Our patient presented with clinical symptoms of cerebrovascular accident. The neck pain which was reported to be present in 71 to 100 percents of patients with cervical vertebral osteomyelitis was not the presenting symptom in our case.<sup>1</sup> Although neck pain was present for about two months, the symptom was mild and ignored as insignificant by the patient. Absence of fever was another factor in which her condition was erroneously attributed to causes other than infection. The prevalence of febrile symptoms was reported as high as 66% in one study. The mean age of patients with cervical vertebral osteomyelitis ranged from 57 to 60 years and majority of patients were male with a range of 66 to 87 percent for male sex.<sup>1</sup> Our patient was an older female patient 72 years of age. She also had no comorbidities or serious medical illness which was reported to be present in 43 to 64 percent of cases.<sup>1</sup> All these factors had contributed in the delay of the diagnosis in this case.

The diagnostic imaging reported in previous case series are mainly based on plain x-ray films and MR imaging in about 55% of patients.<sup>2</sup> Spinal MRI is the most sensitive and specific method of detection with a sensitivity of 93% -96% and a specificity of 92.5% -97%.<sup>3</sup> Our patient had characteristic imaging findings with involvement of two (or more) contiguous vertebral bodies and its intervening disc space, destructive and expansile lesions with low-intensity changes in infected bone and discs on T1-weighted images, high-intensity changes on T2-weighted images and enhancement of involved structures with gadolinium injection.<sup>4</sup> Para-vertebral infection, collections under the longitudinal ligament, and epidural abscesses were also seen.<sup>5</sup> These correspond with previous reported series of high correlation of neurological deficit with the presence of epidural abscess.<sup>6</sup> However, isolated involvement of C1 and C2 vertebrae is rare. The involvement of C5 and C6 were more common (40%), followed by C6-C7 (20%) vertebrae.<sup>1</sup>

Most cases of cervical osteomyelitis are due to haematogenous spread of pyogenic micro-organisms from a distant infective source or following invasive surgical procedures.<sup>7,8</sup> Direct inoculation from adjacent structures is a rarer cause. In our case, the source of infection was not confirmed.

Surgical intervention is indicated in this patient as she had symptomatic neurologic deficit with its complication. Various surgical approaches have been described but anterior debridement and grafting followed by posterior instrumentation has become the mainstay of surgical management. Surgically treated patients generally had favorable outcomes with improvement in neurological function occurring in 66 to 83% of cases.<sup>3</sup> Risk factors for poor outcome include longer time to diagnosis, neurological impairment and secondary sepsis. All of these factors were present in our patient. The mortality rate for osteomyelitis ranges from 2-12%.<sup>9</sup>

## CONCLUSION

This case illustrates the difficulty in establishing early and accurate diagnosis of cervical vertebral osteomyelitis. It remains as a major challenge and heightened awareness about this condition hopefully can avoid diagnostic delay for an optimal outcome.

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