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Case Report

Nonoperative management of craniovertebral junction and cutaneous tuberculosis

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Abstract

Background: Craniovertebral junction (CVJ) and cutaneous tuberculosis (TB) are both rare, each occurring in 0.3–1.0% of patients. To our knowledge, there are no existing cases reporting these manifestations of TB simultaneously. We report a case of TB involving the left CVJ as well as the skin, and discuss our management while providing a review of the literature.

Case Description: An adult patient was presented with progressive nocturnal neck pain associated with the development of several skin lesions. Investigations revealed multiple osseous lesions including the left CVJ. Biopsy of the CVJ lesion was unamenable due to proximity of the vertebral artery; therefore, the patient underwent biopsy of the other sites. Histological examination demonstrated features consistent with TB infection and the patient commenced 12 months of standard anti-TB therapy with cervical spine immobilization. At 2-month review, the patient was well with a near-complete resolution of neck pain and cutaneous lesions. Repeat imaging at 6 months follow-up demonstrated a stable C1 lesion with no evidence of instability.

Conclusion: CVJ TB may be treated solely with anti-TB therapy and immobilization to good effect if there is no gross instability or neurological deficit. Similarly, cutaneous TB responds well to standard anti- TB therapy. Our experience suggests that co-existing tuberculous lesions in the CVJ and skin can be simultaneously managed with standard therapy without significant alterations to treatment regimes or prognosis.



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Key Words: Craniovertebral junction, cutaneous tuberculosis, spinal, tuberculosis

INTRODUCTION

Craniovertebral junction (CVJ) tuberculosis (TB) is, especially uncommon and accounts for 0.3–1.0% of spinal TB, as illustrated in a review by Gupta *et al.* which found only 51 cases of CVJ TB from 1978 to 2004.^[4-6,9,10,12] We report our experience in managing a patient with TB involving the left CVJ as well as the skin and discuss our strategies while reviewing the literature.

CASE REPORT

A 31-year-old male of Indian extraction was presented with a 2-month history of progressive nocturnal neck pain

and 8 months of multiple skin lesions, but no neurological deficit. He had visited India 12 months previously and had received routine bacillus Calmette–Guérin vaccinations before immigrating to Australia at 21 years of age.

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Skin examination revealed; a 4.5 cm solid mass overlying the right deltoid, a 1.5 cm mobile mass on the left chin, and a 1.5 cm mobile mass on the dorsum of the left foot. Although he had a positive QuantiFERON Gold assay (titer 11.68 IU/ml), his chest X-ray was normal. Cervical computerized tomography (CT) showed destructive bony changes at the left atlanto-occipital junction (between the left occipital condyle and superior articular facet of the left lateral mass of C1); there was a soft tissue mass between these plus left-sided lateral subluxation of C1–C2 [Figures 1 and 2].

He was immobilized in a halo vest. A CT of the chest, abdomen, and pelvis showed lytic and sclerotic bony lesions in the ribs, pelvis, sacrum, and right acromion [Figure 3]. As cervical CT angiogram precluded a biopsy of the cervical lesions (e.g., proximity of the vertebral artery), biopsies were obtained from the skin lesions (left pelvis and the right acromion). These revealed necrotizing granulomatous inflammatory changes (but



Figure 1: (a) Axial and (b) coronal cervical spine computed tomography (bony windows) demonstrating destructive bony changes centered in the left atlanto-occipital junction between the base of the left occipital condyle and the superior articular facet of the left lateral mass of CI



Figure 3: Coronal computerized tomography chest, abdomen, and pelvis (bony windows) revealing poorly-defined sclerotic lesions in the left half of the SI vertebral body, right iliac body, left inferior pubic ramus, left ischial tuberosity, and in the left iliac body, abutting the sacroiliac joint

no acid-fast bacilli on smear), while the TB polymerase chain reaction (PCR) was positive. Diagnosed with disseminated TB, he commenced a 1-year course of standard anti-tuberculous therapy (ATT): For example, rifampicin, isoniazid, pyrazinamide, and ethambutol; Lowenstein–Jensen culture confirmed mycobacterium TB with full drug sensitivities. At 2-month review, his neck pain and cutaneous lesions had resolved. Spinal immobilization was maintained for 6 months after which cervical CT and flexion-extension X-rays confirmed Cl/C2 stability [Figure 4].

DISCUSSION

Craniovertebral junction tuberculosis

CVJ TB predominantly affects the anterior structures of the vertebrae. The infection destroys bony and ligamentous structures, resulting in neuraxial compression and instability.^[4,6,10,12,16,17] Diagnosis is difficult due to nonspecific features including suboccipital pain, neurological deficit, and CVJ complex instability.^[4,5,11,12] Tissue biopsy is imperative to confirm the diagnosis and obtain drug sensitivities. PCR is used to make a provisional diagnosis due to its high (>90%) sensitivity and specificity.^[6,12]



Figure 2: (a) Axial and (b) Sagittal cervical spine computerized tomography with bony changes as described in Figure 1. The presence of an associated soft tissue mass between these bony changes and left-sided lateral subluxation of C1 with respect to C2 is also appreciated



Figure 4: (a) Flexion and (b) extension cervical spine X-rays at 6-month review demonstrate no spondylolisthesis or vertebral slippage with stable atlanto-dens interval

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Treatment options for CVJ TB are controversial due to the absence of well-established guidelines.^[1,3,12,14,16] In cases with minimal instability or neurological deficit, conservative treatment with empirical ATT for 12–18 months is recommended.^[1,3,6,8,10,12,16] The most common surgical intervention following decompression and/or biopsy is anterior cervical fusion to stabilize the spine.^[6,10,12,13,16,17] The transoral approach is ideal for biopsy of lesions and decompression of the neuraxis as it gives access to the median and both lateral atlanto-axial joints.^[6,7,10] Immobilization of the cervical spine for 3–6 months is recommended to facilitate bony stability as an adjunct to both nonoperative management and surgery.^[1,3,6,7,9,14,17]

Cutaneous tuberculosis

Cutaneous TB is reported in <1% of cases.^[2,8] The three most prevalent types of cutaneous TB are scrofuloderma, vulgaris, and ΤB verrucosa cutis.^[2,8,15,18] lupus Scrofuloderma is a multibacillary form of cutaneous TB. Lesions present as firm, red-brown nodules overlying a bone or joint.^[2,8] Lupus vulgaris is a paucibacillary form of cutaneous TB and is the most common form of cutaneous TB. Lesions occur in normal skin originating from an underlying TB focus and are solitary, reddish-brown nodules with a gelatinous consistency.^[2,8] TB vertucosa cutis is another paucibacillary form of cutaneous TB and occurs after direct inoculation of TB into the skin. It manifests as a purplish plaque that forms exudative fissures.^[8] Isolation on the culture is difficult; PCR analysis, histopathological features, and responsiveness to ATT within 1 month are therefore used for diagnosis.^[8,15,18] Standard treatment with first-line ATT produces favorable results for all forms of cutaneous TB.[8,18]

Case discussion

The site of the lesion was impacted upon our management decisions. Biopsy was unamenable due to the risk of damaging the adjacent vertebral artery and its associated sequelae. The cutaneous lesions in this case were multifocal, solid subcutaneous nodules with no evidence of exudation. This was consistent with the typical features of lupus vulgaris. As there was no overt instability or compression, we elected for biopsy of the other osseous sites as well as the cutaneous lesions and commenced ATT for treatment. These biopsies indeed demonstrated PCR findings consistent with TB and eventually produced a positive culture.

The long-term outcome of nonoperative management of both CVJ and cutaneous TB is excellent.^[1,3,8-10,12,14,16,18] This was consistent with our case as dramatic improvement of all lesions were experienced 1 week after commencing therapy, with resolution at 2-month review and normal quality of life at 12-month review.

In summary, we present a manifestation of extrapulmonary TB presenting with CVJ and cutaneous lesions, both of which are rare occurrences. We concur with the findings in the literature that CVJ TB can be managed nonoperatively, particularly in the absence of cervical instability or neurological deficit.^[1,3,9,10,12,14,17] Our experience suggests that co-existing tuberculous lesions in the skin and other osseous sites can be simultaneously managed with standard ATT without any significant alterations to treatment regimes or prognosis.

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Conflicts of interest

There are no conflicts of interest.

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