Thoracic Spinal Actinomycetoma: An Under-Recognised Masquerader

P Periyasamy, N Sahari, R Sakthiswary

Citation

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Abstract

Actinomycosis (AM) is associated with bone involvement in only 2% of cases and the thoracic vertebrae is an extremely rare site. We describe a case of a 42-year-old gentleman who presented with constitutional symptoms accompanied by progressive spinal cord compression. MRI spine revealed a paraspinal mass extending from T2 to T9 level with extension into the spinal canal. Histopathological examination of the biopsied lesion stained positive for Actinomyces. This case highlights that AM is another great mimicker similar to Tuberculosis, and should be considered as a differential diagnosis when encountered with a paraspinal mass.

INTRODUCTION

Actinomycosis (AM) is a rare chronic, suppurative granulomatous infection which is usually caused by Actinomyces Israelli. It may occur at almost any site of the human body but common sites include the face, neck, abdomen and pelvis [1]. Even in healthy humans, this anaerobic gram positive bacteria live as a commensal organism adjacent to the sites mentioned above. Spinal AM is extremely rare and the majority of reported cases in the past were not associated with neurological deficits. We hereby report a case of thoracic spinal actinomycetoma in a middle-aged man who presented with paraplegia.

CASE REPORT

A 42-year-old Malaysian gentleman with no previous medical illness presented to us a progressive bilateral lower limb weakness associated with urinary retention. These symptoms were associated with loss of appetite and loss of 10 kg body weight over 3 months. There was no fever and rest of the systemic review was unremarkable. On examination, there was no lymphadenopathy. He had minimal dental carries on his lower teeth. Bilaterally there was reduced sensation up to L1 with generalized muscle power of the lower limbs of 0/5 and plantar response was up-going. Examinations of the cardiovascular, respiratory and abdominal system were normal.

Investigations revealed a haemoglobin of 7.3 g/dl which was normochromic normocytic, total white cell count of 18 $X10^9$/L with neutrophils of 85% and of lymphocytes 10%. C- reactive protein was markedly elevated 11 mg/dl (normal <0.5 mg/dL). Mantoux test was negative. Retroviral screening was non reactive. Computer tomography (CT) of the spine showed an ill defined, heterogeneously enhancing paraspinal mass extending from the level of T2 to T9. There was associated widening of the neuroforamina between the level of T3 and T8 and the lesion narrowed the spinal canal (Figure 1). CT guided biopsy of the lesion was done and histopathology showed a strip of fibrocollagenous tissue which was partially infiltrated by mixed chronic, acute inflammatory cells and histiocytes. Bacterial colony of actinomyces was present, surrounded by acute inflammatory cells. There was no granuloma formation or malignant cells seen. Numerous colonies of Gram and Grocott stain positive filamentous microorganisms were identified to be Actinomyces sp.(Figure 2).

The patient was treated with intravenous C penicillin 4 megaunits qid for 1 month followed by tablet penicillin 4g daily for another 6 months. He was scheduled for a spinal laminectomy but the patient refused surgery. During follow up; 3 months after the commencement of antibiotics, he no longer had urinary retention. However, the dense paraplegia remained the same. But at 8 months of follow up, the muscle power of both his lower limbs improved from 0/5 to 3/5. The clinical improvement was consistent with the radiological reduction in the size of the paraspinal mass on the repeat CT of the spine (Figure 1).

Figure 1

Figure 1: CT scan showing bilateral paraspinal mass (arrows) pre and post treatment

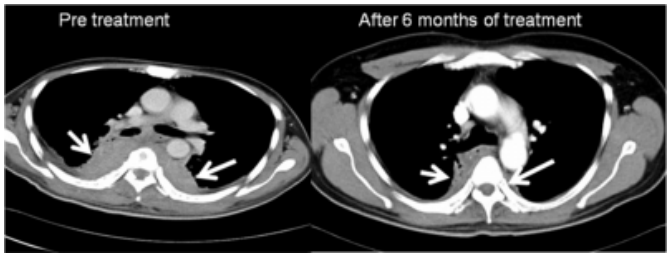
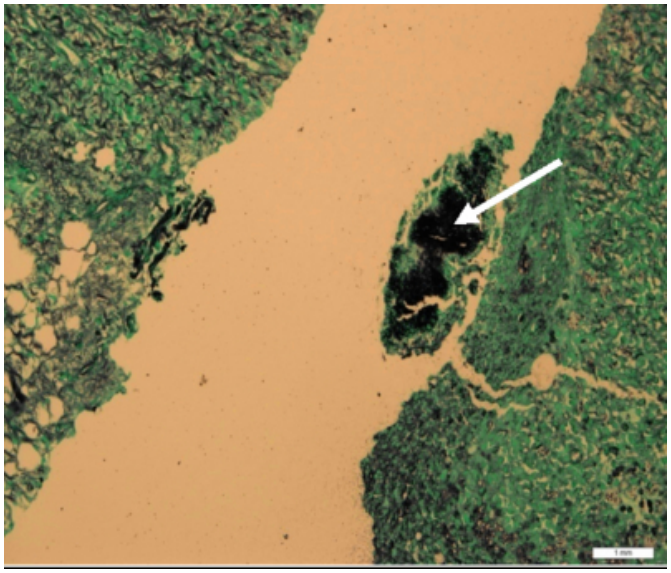


Figure 2

Figure 2: Sulphur granules (arrow) surrounded by inflammatory cells on silver stain.



DISCUSSION

Actinomycosis (AM) is classified as an anaerobic bacterial infection and is not to be mistaken for a fungus as the term 'mycosis' refers to fungal infections. The classification of this microorganism has been controversial since its discovery. It was initially labeled as fungus because of its branching character. This was subsequently revised as a branching gram positive bacteria. Actinomyces sp. is rather unique as it has mixed characteristics of both bacteria and fungus [1].

This case illustrates that AM may occur in an immunocompetent host without any clear cut predisposing factors such as recent dental procedures, poor dentition and alcoholism. Actinomyces have been isolated in 27% of bacteremias associated with dental manipulation [2]. Although AM can occur at virtually any site, involvement of the thoracic vertebrae is extremely rare. We performed a Pubmed search of published articles on AM of the thoracic spine. The keywords used to retrieve the articles were 'actinomycosis', 'thoracic spine' and 'thoracic vertebra'. Our literature search revealed that there were 18 articles on this topic; vast majority of which were case reports. The earliest was in year 1953 while the most recent was in 2008. Most reported cases were from Europe and North America. The cause of the lower incidence in tropical countries is unknown but could be due to underreporting.

Due to the rarity of this infection in the paraspinal region, the diagnosis was not suspected at the time of presentation in almost all previous case reports. Furthermore, AM has non-specific clinical symptoms and signs and has earned a reputation of being a' great pretender 'or 'great masquerader' of chronic conditions such as tuberculosis and malignancies [3]. In most instances, the diagnosis was made either post operatively or following a diagnostic biopsy of the lesion. To date, the only confirmatory investigation is the histopathological examination revealing sulfur granules and branching beaded gram-positive rods in the tissue [4].

It is disappointing that there are no other diagnostic approaches to establish the diagnosis more promptly. Although Polymerase Chain Reaction (PCR) techniques have been developed for actinomycetes, no univocal or standardized protocol exists. PCR has only been experimented in research studies and has not become part of routine clinical practice. Serology has a limited role in this context and remains underexplored. Newer diagnostic tools for AM will hopefully be part of future research agenda in the field of bacteriology.

Researchers, however, have identified certain relatively unique and distinct radiological findings of AM. Cope noted that unlike tuberculosis, there is simultaneous new bone formation of the vertebrae in AM [5]. Brown concurred with the above findings and further described the lesions in AM as spheroidal with surrounding reactive new bone, apparently formed by calcification in the fibrous lining of the cystic areas. AM has the tendency to erode cortical, trabecular and even the subchondral bone. Spinal AM behaves more like fungal infections. But mass-induced erosions in fungal infections are perforated by spicules of new bone which helps distinguish them apart from AM. Indeed, the osseous lesions of tuberculosis with less bony resorption are quite different from the spheroid lesions of actinomycosis [1,6,7]. Moreover, AM typically spares the intervertebral disc [8]. Although our knowledge on the radiological aspect of AM has expanded over the recent

years, it is noteworthy that imaging is a useful guide but the findings per se may not pinpoint to the ultimate diagnosis of AM.

Penicillin is well established as the first line therapy in AM. In instances of penicillin allergy, either doxycycline or sulfonamides such as sulfamethoxazole may be used as an alternative regimen [9]. Most patients respond remarkably to penicillin and it has remained as the anchor drug for decades. This has probably discouraged researchers and clinicians from exploring the use of new antimicrobials in AM. There are no standard international guidelines in this regard. Hence, the basis for current practice is years of clinical experience of experts of the field rather than randomised controlled trials. Prognosis is generally favorable in the vast majority of patients but it may take months to achieve significant clinical improvement as illustrated by this case [10].

CONCLUSION

The diagnosis of thoracic vertebral AM requires a high index of clinical suspicion. It may occur in healthy individuals with no risk factors. Guided by the imaging findings, a microbiologist should be alerted to examine the biopsied specimen promptly for early confirmation of the diagnosis and institution of appropriate anti microbial therapy.

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Author Information

Petrick Periyasamy
Department of Medicine, Universiti Kebangsaan Malaysia Medical Centre

Narisa Sulaiman Sahari
Department of Medicine, Universiti Kebangsaan Malaysia Medical Centre

Rajalingham Sakthiswary
Department of Medicine, Universiti Kebangsaan Malaysia Medical Centre