Epidural Extension Of Actinomycosis- Correct Diagnosis Can Avoid Surgical Intervention: A Case Report

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Citation

Abstract
Vertebral involvement in actinomycosis is very uncommon and epidural extension rare. We report a case of thirty years old HIV infected immunocompromised man who presented with cervical actinomycosis involving cervicodorsal vertebrae with epidural extension causing paraparesis. Patient recovered on long term penicillin treatment without any surgical intervention. Though actinomycosis is uncommon in this era, when present, it poses a diagnostic challenge. Hence it is worthwhile to consider this possibility in any soft tissue tumoral formation on the face and neck, especially in presence of discharging sinuses. Appropriate use of imaging and laboratory investigations aids in prompt and accurate diagnosis of this difficult condition and obviates surgical treatment. Tuberculosis is the closest differential diagnosis in such cases. Differentiating features from tuberculosis are also considered.

Vertebral involvement in actinomycosis is very uncommon. We report a case of cervical actinomycosis involving cervical vertebrae with epidural extension leading to paraparesis. Less than 20 cases of actinomycosis-related spinal neurological deficit have been documented. Actinomycosis is usually mistaken for tuberculosis.

CASE HISTORY
A thirty years old HIV infected immunocompromised gentleman presented with progressive paraparesis and acute onset severe radicular pain and tingling in both upper limbs since four to five days. He had a history of swelling over back of neck since four years for which he had sought local treatment in his village. He denied any history of fall, lifting weights or fever. On examination the patient was emaciated, had severe neck stiffness and grade I power with sensory loss in lower limbs. Loss of sensation was also noted below mid-thoracic level. A large erythematous indurated swelling over nape of neck and upper back, extending up to left shoulder. Multiple discharging sinuses and few depressed scars were present. Multiple tiny yellowish granules were present in the pus. Few enlarged cervical lymph nodes were present. Routine lab investigations were normal apart from mild anemia and raised ESR (106 at the end of first hour). Considering the discharging sinuses and presence of granules, actinomycosis or nocardiosis was considered to be one of the diagnostic possibilities.

Radiograph (Figure 1) of cervical spine showed mild loss of lordosis and presence of permeative lytic areas involving spinous processes of C4-5 vertebrae with widening of prevertebral soft tissue. Posterior cervical soft tissue shadow was also noted. Limited computed tomography sections (Figure 2) confirmed these findings.
Figure 1
Figure 1: Lateral cervical spine radiograph. Note the presence of permeative lytic areas involving spinous processes of C4-5 vertebrae with widening of prevertebral soft tissue and presence of posterior cervical soft tissue shadow.

Figure 2
Figure 2: Computed tomography. Multiple lytic areas seen involving bodies and posterior elements.

Plain and contrast enhanced magnetic resonance imaging (MRI) (Figure 3-6) revealed areas of altered signal intensity in spinous processes, laminae, pedicles, transverse processes and bodies of C4 to D3 vertebrae. Patchy involvement of heads of adjacent ribs was also noted. These areas were iso to hypointense on T1 weighted images, hyperintense on T2 weighted images and some of them showed enhancement following gadolinium injection. Enhancing epidural soft tissue causing cord compression was present, extending from C6 to D3 level. Enhancing prevertebral and paravertebral soft tissue was also present at the same level. Dense infiltrates were noted in apical region of left lung, adjacent to D2-3 vertebrae (not shown). Whole spine screening did not reveal any other abnormality.
Figure 3
Figure 3: MRI T1 weighted sagittal image. Note the areas of altered signal intensity in posterior elements and bodies of C6 to D3 vertebrae. Epidural soft tissue causing cord compression is present, extending from C6 to D3 level. Note also the prevertebral soft tissue.

Figure 4
Figure 4: MRI T2 STIR sagittal image. Note the epidural extension of soft tissue and multilevel bone involvement. Also note D1 vertebral collapse. Involvement of C4-5 and posterior cervical inhomogenous soft tissue is better appreciated here.

Incisional biopsy was performed on arrival. Discharged material (sulphur granules) was also sent for microscopy and culture. Microscopy confirmed presence of actinomycosis. Infiltrates of histiocytes, plasma cells and many lymphocytes, forming local aggregates at places were seen along with multiple actinomycetic granules surrounded by dense polymorphonuclear aggregates. The granules showed central dense basophilic material and a lightly stained peripheral portion with characteristic radiating clubs. Furthermore, they revealed colonies of actinomyces comprised of Gram positive, nonacid fast, slender, branching and beaded organisms.

Urgent decompression was considered, but in view of clinical and imaging findings the patient was started on intravenous penicillin. Surgical intervention would serve as standby treatment in case patient deteriorated. Later on culture confirmed diagnosis of Actinomyces viscosus (after 14 to 16 days).

Patient gradually recovered. After 5 months of treatment, patient’s power in lower limb was grade IV. Only slight limitation of movement was present. Follow up MRI showed considerable improvement. Patchy areas of altered signal intensity were still present in bones.

DISCUSSION
Actinomycosis is an anaerobic gram-positive, branched,
filamentous bacterium, residing as commensal in periodontal pockets and gingival crevices, in carious teeth, dental plaques, tonsillar crypts or in periodontium. Dental extraction and trauma are the most important risk factors for development of actinomycosis.

Cervicofacial region (50-60%), chest and the abdomen are most commonly affected. Other possible sites of infection are the extremities, lacrimal glands, kidneys, genital organs, bones and the central nervous system. Actinomycosis is mostly found in young adults, more often seen in males than in females.

Vertebral involvement occurs invariably by direct extension of the infection from a primary site. Initially there is pre and paravertebral ‘phlegm’ formation. Then the bone is involved with periositis. One or several successive vertebrae may be involved. Sometimes new bone formation may be seen, giving mottled appearance on radiographs. As the disease progresses, the initial erosions become deeper causing destruction of the cancellous bone of the vertebra with secondary osteomyelitis, and abscess formation resulting in multiple intercommunicating cystic spaces. Sequestrum formation does not occur.

Presentation may be nonspecific, with fever, weight loss, fatigue. Nodular lesions may be present that increase in size and number, ultimately forming discharging sinuses. Sulfur granules may be seen in the exudate.

Tuberculosis is the closest differential diagnosis. In tuberculosis, unlike actinomycosis, in tuberculosis, the initial focus starts by haematogenous spread to the cortex of the bone. Actinomycosis usually spares the discs, but unlike tuberculosis invariably affects the adjacent pedicles, transverse processes and corresponding heads of the ribs [\textsuperscript{6}]. As a result, vertebral collapse and angular deformities are uncommon [\textsuperscript{6}]. However vertebral collapse may be present in aggressive form of actinomycosis. Metastatic disease is another differential diagnosis [\textsuperscript{6}].

Penicillin is the drug of choice. Tetracycline may also be used. Long term treatment is needed. Though prompt surgical decompression is advocated [\textsuperscript{7}], it may be reserved as a second line treatment if the condition is diagnosed promptly and appropriately treated. External fixation can be used for stabilization of the spine in salvage cases. It avoids the need for internal fixation devices and for postoperative bracing and is also reported to have no major complications [\textsuperscript{7}].

CONCLUSION

Though actinomycosis is uncommon in this era, when present, it presents a diagnostic challenge. Hence it is worthwhile to consider possibility of actinomycosis in any soft tissue tumoral formation on the face and neck, especially in presence of discharging sinuses. Appropriate use of imaging and laboratory investigations aids in prompt and accurate diagnosis of this difficult condition and may obviate need of surgical treatment. CT guided aspiration may be useful in obtaining tissue sample for diagnosis.

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