CASE REPORT

Sporadic actinomycosis of the hip complicated by Central Nervous System infection

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Abstract

Actinomycosis is caused by the Gram positive filamentous Actinomyces bacterial species that are normal commensals of the oral cavity. Due to their low virulence, disease is rare in the immune competent patient. Although it may afflict any system in the body, involvement of the musculoskeletal system is uncommon. Here in, we describe the case of a 60 year old lady presenting with low grade fever, left hip pain and drowsiness. She was diagnosed as left hip actinomycosis on Computed tomogram (CT) guided biopsy and histopathological analysis of infiltrative lesions identified on Magnetic Resonance Imaging (MRI). She also had meningitis diagnosed on cerebrospinal fluid analysis which improved with treatment of actinomycosis. Actinomycosis of the hip is rare, and occurs in the presence of described predisposing factors. To the best of our knowledge, this is the first case of sporadic actinomycosis of the hip complicated by meningitis in an immune competent individual.

Keywords: Actinomycosis, Meningitis, Immune competent.

Introduction

Actinomyces have traditionally been classified as bacteria although they have some homology to fungi.[1,2] These filamentous gram positive bacteria produce multiple abscesses and sinus tracts often producing a yellowish discharge termed "sulfur granules".[2] Actinomycosis can affect many organ systems. The most common type is cervico facial disease. Involvement of the abdomen, pelvis and respiratory tract account for the remaining cases.[3] In the pelvis, bony structures may be involved. Actinomycosis of the native hip is a rare entity. We performed a thorough PubMed search and found only 8 cases of hip actinomycosis. Predispositions identified were prosthetic hip joints, immune compromise due to various reasons, post surgical procedures, trauma, diabetes mellitus or intravenous (IV) drug abuse.[4]

We describe the case of a sixty year old lady, who presented with unremitting pyrexia, gradually worsening left hip pain and drowsiness. To the best of our knowledge, this is the first case of actinomycosis of the native hip joint in an immune-competent patient without known predisposing factors. It also describes the unusual dissemination of the infection to the central nervous system (CNS).

Case Report

A 60 years old hypertensive lady with no other co morbidities presented to the Department of Internal Medicine, Shifa International Hospital Islamabad in November 2015 with left hip pain and fever for the past 3 months, and progressive drowsiness for three days. The pain was localized to the joint, gradual in onset, exacerbated by movement and relieved by analgesics. The fever was low grade, intermittent and responded to anti pyretics.

On examination she was a middle aged lady with pulse rate 105/minute, blood pressure 110/70 mm hg, respiratory rate 18/minute and temperature 100°F. Her oral hygiene was satisfactory. She had poor skin turgor and dry mucous membranes. Examinations of the cardiovascular, gastro intestinal and respiratory systems were unremarkable. Neurological examination revealed a Glasgow comma scale (GCS) of 13/15. There was mild neck stiffness but no other signs of meningism. Other components of the examination were unremarkable. Local examination of the left hip revealed mild tenderness to deep palpation.

A radiograph of the left hip showed decreased bone density and joint space (Figure: panel A). Pertinent lab investigations had a total leukocyte count of 16,400/cumm (4000-11000/cumm), Erythrocyte sedimentation rate 60 mm/1st hour (0-20 mm/1st hour), serum sodium 121 (135-145 meq/L), C reactive Protein (CRP) 309 mg/dL (less than 5 mg/dL). Numerous white blood cells and positive leukocyte esterase were seen in routine urine examination. Initial cerebrospinal fluid (CSF) analysis is tabulated in Table. Measured serum osmolality was 270 mOsm/kg/L (285-295 mOsm/kg/L), urine

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Table-1: A. Comparison of the Cerebrospinal Fluid (CSF) analysis before and after antibiotic therapy. B. A comparison of the magnetic resonance image (MRI) diameters before and after antibiotic therapy.

A. CSF Parameters	Pre-treatment (Ampicillin)	Post-treatment (Ampicillin)	Reference Range
Appearance	Clear	Clear	Clear
White Blood Cells (counted on chamber)	33	10	0-5 cells/mm^3
Neutrophils	40%	15%	0%
Lymphocytes	60%	85%	100%
Red Blood Cells (counted on chamber)	05	20	0-10 cells/mm^3
Proteins	76.4	50.3	15-45 mg/dl
Glucose	41	67	50-75 mg/dl
Lactate Dehydrogenase	40	39	< or = 40 units/l

B. MRI Diameters	Pre-treatment (Ampicillin)	Post-treatment (Ampicillin)
Oblique Antero-posterior (AP)	7.3 centimeters	3.3 centimeters
Oblique Transverse (T)	18 centimeters	14 centimeters
Cranio-caudal (CC)	17 centimeters	13 centimeters

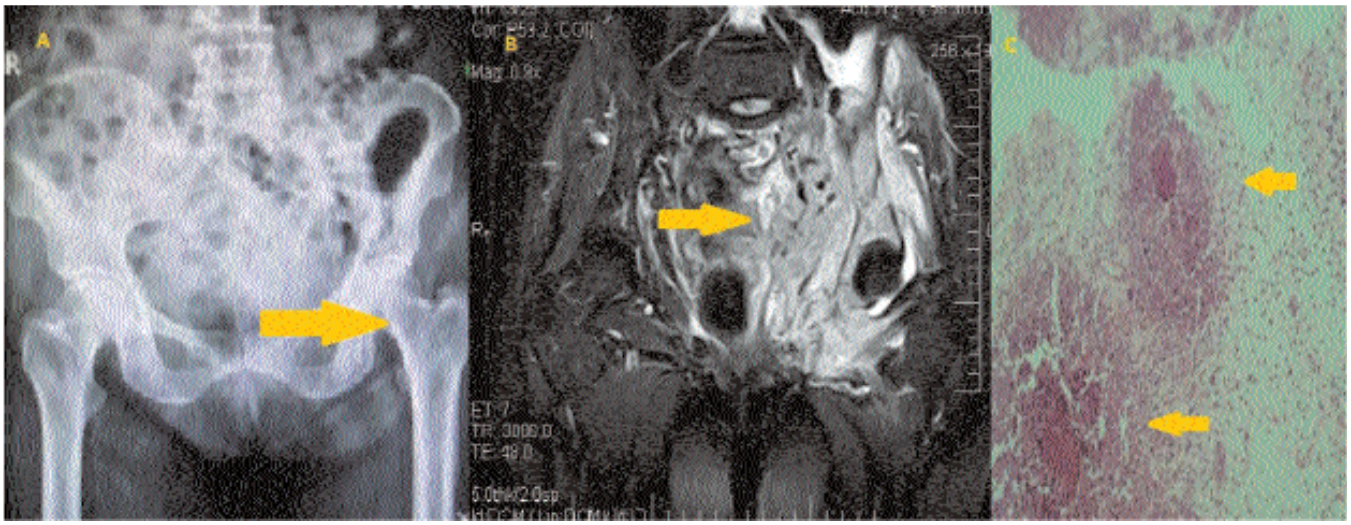

Figure-1: Panel A) A radiograph of the hip revealing severe osteopenia of the left hip, femoral head, neck and shaft. Panel B) Magnetic Resonance Image (MRI) showing a large infitrative lesion involving the left hip joint. Panel C) Histopathology image of the biopsied tissue showing.

osmolality 320 mOsm/Kg/L. Random blood sugar was 100 mg/dL and HbA1c was 5.8% (<6.5 %)

Magnetic resonance image (MRI) of left hip revealed large infiltrative lesion involving the left hip bone mimicking an osteosarcoma (Figure: Panel B). A computed tomogram (CT) guided biopsy of the lesion was performed for histo pathological/microbiological analysis.

Despite being treated for 2 days with intravenous ceftriaxone and normal saline in a high dependency unit, she developed progressive obtundation and needed ventilator support. Blood and urine cultures did not yield any pathogen. The serum sodium rose to 125 meq / L and 130 meq/ L in the following 2 days. The hip biopsy report showed multiple colonies of actinomyces israelii seen as filamentous basophilic bacteria arranged in rosettes surrounded by inflammatory cells and granulomatous reaction (Figure: D). Intravenous Ampicillin was initiated at a dose of 1 gram q 6 hourly. The response was dramatic, with a fall in the inflammatory markers, improvement in the conscious status followed by extubation. A repeat CSF analysis showed resolving infection as illustrated as a comparison in Table. A repeat Magnetic Resonance Image (MRI) one week after initiating therapy confirmed lesion size reduction. This has been

tabulated as a comparison in Table.

Infectious disease consultant advised Intravenous treatment for 6 weeks followed by amoxicillin 500mg orally thrice daily for 6 months. Currently, the patient is doing well, complying with therapy and visiting follow up clinics regularly.

Discussion

More than 30 species of actinomyces exist which can target almost any site in the body. The commonest specie is actinomyces Israelii.[5] This was also the causative agent in our case. The organism is slow growing and lesions frequently mimic chronic conditions like tuberculosis, nocardiosis and even malignancy; some cases may be complicated by cold abscesses.[5,6]

Orocervicofacial disease is the commonest type of actinomyocis.[7,8] Other sites of involvement include the thoracic, abdominal and pelvic regions. Musculoskeletal disease is relatively uncommon. This may occur via contiguous or haematogenous spread from an actinomycotic focus.[6] Although disease involving the spine has been reported more commonly with A. Israelii and A. meyeri, involvement of the hip joint is a rare entity. Predispositions include trauma to the hip, immune suppression, prosthetic hip joint replacement, diabetes mellitus and intravenous drug use.[9,10] Past reports also mention poor dental hygiene and dentition as predisposing factors.[8] Our case was special since our patient had no predisposing factors for the development of actinomycosis of the hip. This was confirmed by a thorough history, clinical examinations and lab analysis for the above mentioned predispositions. No other primary focus of the disease was identified.

Diagnosing actinomycosis can be a dilemma since it mimics other chronic and slow growing lesions. Initial diagnostic modalities include CT and MRI for the localization and extent of the lesions. Our case was reported by the radiologist with an initial suspicion of an osteogenic cancer. The best test to formally diagnose the disease is via tissue sampling/biopsy with histo pathological analysis. Our case was diagnosed by this method. This involves gram staining of the collected pus/tissue obtained followed by microscopic examination; typical microscopic findings include necrosis with yellowish sulfur granules and filamentous Gram-positive fungal-like filamentous and branching pathogens. Our findings were similar. Cultures are sterile in 50% of the cases, mostly since actinomyces grow in anaerobic conditions and require special precautions for isolation.[6] Our cultures were also inconclusive.

The CNS is frequently involved by actinomycosis with brain abscesses the most common pathology. Other manifestations include meningitis, encephalitis, epidural abscess and subdural empyema. The involvement typically occurs either haematogenously from the respiratory tract, direct extension from cervico facial disease or after a penetrating head injury.[6] Systemic spread from a primary hip joint infection has never been described before, and we believe that this was the primary focus that disseminated causing meningitis. CSF findings include a polymorphonuclear leukocytosis, elevated protein and normal or low glucose levels.[7] Our patient demonstrated a similar lab analysis. Contrary to our case, more commonly CNS disease is associated with A.meyeri infection.[7]

Treatment involves using antimicrobial therapy at high doses for prolonged duration. Drug resistance is currently not a problem. High doses of penicillin G, amoxicillin or ampicilin may be used for first line treatment.[6] The duration of therapy may be as much as 6-12 months depending upon the clinical response.[6] Our patient was given intravenous ampicilin therapy for 6 weeks followed by a six month course of amoxicillin. The response was excellent with good compliance. Surgery has been used routinely in the past for actinomycotic lesions. Current trends however, advocate, limiting invasive procedures and focusing on a targeted antibiotic regimen. Abscesses are drained and resective surgery may be employed if drug therapy fails or extensive necrosis in encountered.[8]

Conclusion

Actinomycosis of the hip may occur in immune competent individuals with no described predispositions. The disease may disseminate to cause CNS infection. Early diagnosis with tissue sampling and treatment with high dose penicillin is pivotal for reducing morbidity and mortality.

Disclaimer: The manuscript was presented as a poster at Annual Research day at Shifa International Hospital Held on 19-20th December abstract book poster 20, Islamabad, Pakistan where it won first position.

Consent and Institute Approval: Informed consent was sought from the patient and her family for the drafting and publishing of her case. Approval has also been taken from the Institutional Review Board (IRB) and ethics

committee of Shifa International Hospital, for the publishing of this case.

Conflict of Interest: None.

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