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Case Report

An unusual case of fungal rib osteomyelitis with sequestrum extending into the segmental bronchus in an immunocompetent host

Neha Bagri[1]*, **Arthi Yadav**[1], **Atha Ulla**[1], **Santosh Sadafule**[1]

[1]*Dept. of Radio-Diagnosis, VMMC & Safdarjung Hospital, New Delhi, India*

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ABSTRACT

Osteomyelitis of rib is rare and mostly occurs in children. In our country, an important cause of rib osteomyelitis is tubercular infection from adjacent lung, lymph nodes or from vertebra. Fungus is a rare cause of osteomyelitis, with organism responsible being, Candida, Aspergillus etc. It is usually associated with immunosuppressive states. We hereby report a case of 36-year-old male who presented with complaints of cough and expectoration of thick mucoid sputum for 1 year, diagnosed as fungal pneumonia 6 months back, with patient not improving after antifungal therapy. On follow up imaging, this patient showed an unusual presentation of rib osteomyelitis with sequestrum extending intrabronchial into inferior segmental bronchus.

1. Introduction

Osteomyelitis of the rib is a rare occurrence. Staphylococcus aureus is the most common organism responsible for rib osteomyelitis, although atypical organisms have also been implicated.[1] However, the most common cause of rib osteomyelitis in our country is tubercular infection, secondary to primary infection of the lung, lymph nodes or from vertebra. Fungal osteomyelitis usually occurs in immunosuppressive patients, with Candida being most commonly responsible. Aspergillus is a saprophytic fungus which is found ubiquitously in environment and commonly affects immunocompromised hosts.[2] There are various manifestations of osteomyelitis on conventional radiography, with the first bony changes on radiography occurring after 2 to 3 weeks. MRI also plays an important role in demonstrating the associated findings and complications of osteomyelitis, with the usual findings being bone marrow oedema, sequestrum, involucrum, sinus tracts formation etc. The present case report shows an unusual and rare presentation of rib osteomyelitis with the sequestrum of the rib seen extending into the segmental bronchus of the adjacent lung, occurring as a complication of fungal pneumonia in an immunocompetent patient. There are no similar case reports with this unusual presentation available in the literature.

2. Case Report

2.1. Clinical presentation

A 36-year-old male presented with complaints of cough and expectoration of thick mucoid sputum for 1 year. He had history of episodic haemoptysis and weight loss (10 kgs in last 6 months). Microscopy for sputum AFB was done which was negative. KOH mount showed thin hyphae in the mucus strands. BAL for galactomannan was positive. However, cytopathology report showed few neutrophils and pigment laden macrophages, but no granuloma or fungal hyphae was seen. Contrast enhanced CT (CECT) was done 6 months back which showed left lower lobe consolidation and mediastinal lymphadenopathy. Patient was started on

* *Corresponding author*.
E-mail address: drnehabagri@gmail.com (N. Bagri).

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itraconazole, however patient continued to have cough with expectoration. Patient did not have any pain, swelling or discharge from the chest wall. Patient was a non-smoker and denied alcohol consumption.

2.2. Imaging findings

Chest x ray showed lytic lesion in the left 8^{th} rib with slight expansion of the rib (Figure 1). CECT revealed evidence of cortical destruction and sclerosis of body of the left 8^{th} rib (predominantly on the medial aspect), with adjacent enhancing soft tissue component and extra-pleural fat proliferation suggesting chronic osteomyelitis. There was also break in continuity of cortex, suggesting pathological fracture. A thin, linear hyperdense fragment (1208 HU), measuring 2.3 x 0.3 cm was seen extending from the bony crater of left 8^{th} rib to the segmental inferior lingula bronchi and lateral basal segment of left lower lobe, likely sequestrum (Figure 2). There was also associated area of fibrosis and tractional bronchiectasis. Varicoid bronchiectatic changes were also seen in posterior basal segments of left lower lobe with adjacent subpleural cyst formation.

Figure 1: Shows expansile lytic lesion of the left 8^{th} rib (arrow)

CEMRI of the chest wall was done which showed area of altered signal intensity involving the body of left 8^{th} rib with T1 isointense and T2 hyperintense soft tissue seen along the medial border of the rib of approximate length 3.2 x 0.8 cm causing medial cortical breach. This soft tissue showed homogenous contrast enhancement on post contrast sequence (Figure 3). There was extra-pleural fat proliferation with enhancing collapsed adjacent subsegmental lung. On GRE sequence, the central area appeared hypointense extending from the rib into the adjacent lung (approximate length 2.2 cm) suggestive of bone or calcification. There was cortical T2 hypo-intensity around the above-mentioned cortical breach suggestive of sclerosis of adjacent bone. On DWI, there was no evidence of diffusion restriction.

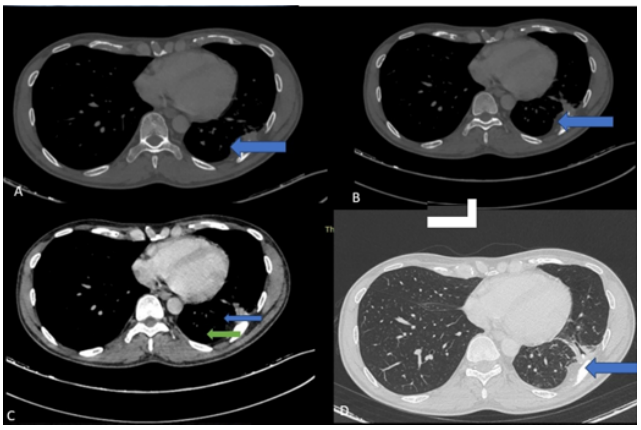

Figure 2: A-D: A: Shows cortical destruction and sclerosis of body of the left 8^{th} rib on the medial aspect (arrow), **B:** shows thin break in the continuity of cortex (arrow) suggesting pathological fracture, **C:** Shows enhancing soft tissue component adjacent to the left 8^{th} rib (blue arrow) and extra-pleural fat proliferation (green arrow), **D:** Shows thin, linear hyperdense fragment (arrow) extending from the bony crater of left 8^{th} rib to the segmental inferior lingular bronchi, likely sequestrum

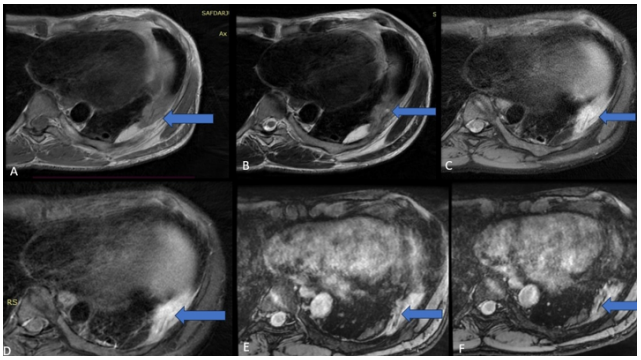

Figure 3: A-F: A (T1 W sequence), **B:** (T2 W sequence), **C:** (T1 FS) and **D:** (T1FS Post contrast) shows altered signal intensity of body of left 8^{th} rib with T1 isointense, T2 slightly hyperintense and T1FS hyperintense soft tissue (arrows) along its medial border causing medial cortical breach. **E** and **F:** (GRE) shows central area appearing hypointense extending from the rib into the adjacent lung suggestive of bone or calcification (in this case sequestrum).

3. Discussion

Fungal pneumonia and invasive fungal infections lead to substantial morbidity and mortality in immunosuppressed patients, however is less common in immunocompetent patients. Fungal infections can lead to various complications like sinusitis, meningitis, osteomyelitis etc. Osteomyelitis of rib is one such rare complication of fungal pneumonia.[3]

Table 1: Differential diagnosis of Rib osteomyelitis.

	Osteomyelitis	**Reactive osteitis**	**Malignancy (chest wall Ewings sarcoma)**	**Stress injuries**
Clinical features	Occurs as a sequela to infection, which leads to destruction of the bone with involucrum, sequestrum, sinus, intraosseous abscess formation.	Occurs secondary to trauma, surrounding infections etc. Usually does not lead to involucrum, sequestrum or intraosseous abscess formation.	Usually has a much slower course compared osteomyelitis, which causes a rapid destructive change	Bones which undergo repetitive stress leads to stress injuries
Imaging findings	Well defined area of low signal intensity on T1, usually **intramedullary distribution.** Non enhancing sequestrum which is low signal intensity on T1, T2 and STIR and enhancing granulation tissue which is intermediate to low signal intensity on T1 and high signal intensity on T2/ STIR sequences.	Ill-defined area of intermediate or low signal intensity on T1 in a **subcortical distribution**.	Destruction of the involved bone (rib) with surrounding soft tissue proliferation, chondroid matrix formation and invasion into the surrounding visceral structures	Leads to marrow oedema with periosteal reaction similar to osteomyelitis. But, the signal abnormalities in stress injuries are confined to bone and there is no inflammatory change in surrounding soft tissues.

However, in our country tuberculosis is a major health problem and musculoskeletal TB accounts for 1-3 % all cases of TB and 15 % of extrapulmonary TB. Vertebral involvement is the most common form of skeletal TB, however, other less common sites of skeletal involvement include hip, knee, foot, elbow, clavicle, sternum, ribs etc.[4]

The first manifestation of osteomyelitis on conventional radiography are soft tissue changes muscle swelling with loss of fat planes. Bony changes are seen after 2 to 3 weeks, which include periosteal thickening, endosteal scalloping, lytic lesions, osteopenia, and loss of trabecular architecture. Acute osteomyelitis if not treated can develop into subacute osteomyelitis (Brodie's abscess) which is sharply delineated focus of infection surrounded by granulation tissue and chronic osteomyelitis which leads to bony erosion, sequestrum (necrotic bone), involucrum formation, chronic discharging sinuses, subperiosteal abscess with lifting of periosteum, soft tissue fistula etc. CT shows abnormal thickening of the cortical bone with sclerosis, sequestrum and involucrum formation, cloacas, intraosseous gas etc.[3] MRI plays an important role in the diagnosis of osteomyelitis as it has excellent spatial resolution.[4] It allows early detection of osteomyelitis as early as 3 to 5 days after onset of infection.[5]The earliest finding on MRI includes alteration in bone marrow signal intensity and marrow oedema which appears as ill-defined low signal intensity on T1 and high signal intensity on T2 and fat suppressed sequences and does not enhance on contrast injection. Sequestrum is seen as low signal intensity on T1, T2 and STIR, which is non enhancing, whereas the granulation tissue is seen as intermediate to low signal intensity on T1 and high signal intensity on T2 or STIR sequences and enhances on contrast administration.[6] A cloaca is seen as a high signal intensity gap in the involucrum on T2 sequences.[7] Tubercular involvement of the ribs also presents as bony erosions with disruption cortical margins and abscess formation. Tubercular abscesses can be formed which can involve the chest wall, sternum, costochondral joints, ribs, vertebrae etc.[8] Intraosseous abscess and subperiosteal abscess are also complications of osteomyelitis, especially tubercular osteomyelitis. They are seen on MRI as low signal intensity on T1, high signal intensity on T2 and shows peripheral contrast enhancement.[9] Bone Scintigraphy can also be used for the diagnosis of osteomyelitis using agents such as ^{99m}Tc MDP, ^{111}In labelled leukocyte and Gallium-67. Bone scan shows abnormal uptake of radionucleotide 10 to 14 days before changes of bone mineral density are seen on plain radiographs. Bone scan is done in three phases and osteomyelitis shows focally increased uptake in all the three phases.[10] Lastly, the gold standard for the diagnosis of osteomyelitis is bone biopsy with histopathological examination and tissue culture.[11]

The present case is an unusual presentation of rib osteomyelitis which occurred as a complication of fungal pneumonia. In the present case, there was lytic lesion in the left 8^{th} rib which showed cortical destruction with sclerosis of body of the left 8^{th} rib, enhancing soft tissue component and a linear hyperdense fragment extending from the bony crater of left 8^{th} rib to the segmental inferior lingula bronchi, likely sequestrum on CT and an area of altered signal intensity involving the body of left 8^{th} rib with T1 isointense and T2 hyperintense enhancing soft tissue on MRI with a central linear hypointense area on GRE

extending from the rib into the adjacent lung suggestive of bone or calcification, which was the sequestrum in our case.

4. Differential Diagnosis

Differential diagnosis of rib osteomyelitis includes reactive osteitis, malignancy (chest wall Ewings sarcoma), stress injuries, chest wall abscess including subcutaneous abscess, inter or intramuscular abscess, etc. Osteomyelitis presents as an area of altered signal intensity in the medullary cavity with enhancing granulation tissue and non-enhancing sequestrum. Reactive osteitis occurs most commonly after trauma or surrounding infections, usually presents as ill-defined area of altered signal intensity in subcortical distribution and does not lead to involucrum, sequestrum or intraosseous abscess formation. Malignancy has a much slower course of presentation compared osteomyelitis, which causes a rapid destructive change. There is destruction of the involved bone with soft tissue proliferation, matrix formation, periosteal reaction, invasion into the surrounding structures and metastasis. Stress fractures occurs after repetitive trauma and the signal abnormalities in stress injuries are confined to the bone with no inflammatory changes in surrounding soft tissues. Chest wall abscess including subcutaneous abscess, inter or intramuscular abscess occurs following tubercular infections and leads to formation of well-defined peripherally enhancing collections in the soft tissue with minimal changes in the surrounding bones.

The distinguishing features are described in Table 1.

5. Conclusion

Rib osteomyelitis is an uncommon complication of fungal pneumonia, especially in immunocompetent patients. Osteomyelitis leads to various imaging manifestations on radiography like soft tissue swelling with obliteration of soft tissue plane, periostitis, cortical lucency, involucrum, sequestrum formation, sinus tracts etc. MRI plays an important role as it has excellent spatial resolution for early detection of osteomyelitis. There is alteration in bone marrow signal intensity, marrow oedema, enhancing granulation tissue, involucrum, cloaca and sequestrum formation. There can be other findings such as disruption cortical margins and abscess formation, especially in patients with tubercular osteomyelitis. The present case report shows an unusual presentation of rib osteomyelitis in a patient presented with fungal pneumonia 6 months back. CECT was done which showed left lower lobe consolidation and mediastinal lymphadenopathy. On follow up CT, there was cortical destruction and sclerosis of body of the left 8^{th} rib, with a linear hyperdense fragment (1208 HU) extending from the bony crater of left 8^{th} rib to the segmental inferior lingular bronchi. On MRI, this fragment was seen as central linear area of blooming on GRE sequence, likely representing the sequestrum of chronic osteomyelitis, because of which the patient continued to have to have cough with expectoration even after itraconazole therapy. Such presentation is a rare event and should be kept in mind while dealing with a case of osteomyelitis of the rib.

5.1. Teaching Points

1. Rib osteomyelitis is an uncommon complication of fungal pneumonia. Various imaging manifestations on radiography include soft tissue swelling with obliteration of soft tissue plane, periosteitis, cortical lucency, involucrum, sequestrum formation. On MRI, there is marrow oedema, enhancing granulation tissue, involucrum, cloaca and sequestrum formation.
2. MRI plays an important role in diagnosing rare complications of osteomyelitis and also helps us in differentiating osteomyelitis from other conditions like reactive osteitis, stress fractures, malignancy etc.

5.1.1. MCQs

1. What is the earliest finding of osteomyelitis on radiograph?

   (a) Periosteal thickening
   (b) Endosteal scalloping
   (c) Loss of soft tissue fat planes
   (d) Lytic lesion
   Answer: C

2. Which of the following imaging modality allows earliest detection of osteomyelitis?

   (a) X-ray
   (b) CT
   (c) USG
   (d) MRI
   Answer: D

3. Which of the following is not an imaging manifestation of osteomyelitis?

   (a) Involucrum
   (b) Cloaca
   (c) Subperiosteal abscess
   (d) Matrix mineralization
   Answer: D

6. Conflict of Interest

None.

7. Source of Funding

None.

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Author's biography

Neha Bagri, Associate Professor https://orcid.org/0000-0002-4883-1300

Arthi Yadav, Junior Resident

Atha Ulla, Senior Resident

Santosh Sadafule, Junior Resident

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