An Unusual Differential Diagnosis of Sarcoma

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Abstract

Osteomyelitis is an uncommon cause of a soft tissue mass and is diagnosed clinically by a blend of radiology, histology, and microbiology. This case report outlines a case of melioidotic osteomyelitis presenting as a suspected sarcoma. The diagnosis of melioidotic osteomyelitis was done against the diagnosis of sarcoma only after radiological evaluation, microbiological evidence of Burkholeria pseudomallei and osteomyelitis confirmed on histology. It is an extremely rare mimic of primary sarcoma of bone and the presentation described in the present case report appears to be unique.

Introduction

Osteomyelitis is an uncommon cause of sarcoma of bone and is diagnosed clinically by a blend of radiology, histology, and microbiology. Very rarely, osteomyelitis can mimic a primary bone neoplasm [1]. Burkholeria pseudomallei (B. pseudomallei) causes melioidosis, and this is a gram-negative environmental saprophyte very often found in wet soils and contaminated waters of endemic areas [2]. As we know melioidosis is sporadic in many parts of the world but is endemic in Southeast Asian countries, such as India and northern Australia, and some parts of tropical and sub-tropical places [3]. We still know very little about the factors that influence the disease's comprehensive description, clinical course, and presentation. However, variations in the virulence of infecting strains in conjunction with host immune competence are the two major contributing factors to disease progression and prognosis. Here we present a case of a 65-year-old female with a suspected left humerus sarcoma case and how she

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was diagnosed with melioidosis.

**Case presentation**

Our patient was a 65-year-old female from Panvel, Navi Mumbai, Maharashtra was a known case of diabetes mellitus, it was a referral case of left humerus sarcoma with secondaries to both lungs diagnosed on PETCT (Figure 1 and Figure 2), one week ago before referral to our center. She presented in our emergency department with a two-week history of left shoulder pain, fever, breathlessness at rest, bilateral pain, and swelling in both lower limbs. On admission, she was hypotensive with a blood pressure of 70/60mmHg, with a fever of 101°F and 94% oxygen saturation on room air.

![Figure 1: PETCT A. Axial view B. Coronal view showing multiple solid angiocentric pleuro-parenchymal nodules of varying sizes showing increased FGD uptake in both the lungs suggestive of metastatic disease. The SUV max of the reference nodules was 11.66 and measured 1.3x1.11 cms.](image)

![Figure 2: PETCT A. Axial view B. Coronal view showing evidence of hypermetabolic soft tissue lesion surrounding the neck and proximal shaft of humerus and extent involving deltoid group of muscles most likely of neoplastic etiology.](image)
On physical examination, she had reduced air entry in both lung fields and basal crepitations, restricted range of movements in the left shoulder, and increased warmth and swelling over both the lower limbs suggestive of lower limb cellulitis. Routine lab investigations were sent, and blood and urine cultures were sent (Table 1). She started empirical antibiotic therapy.

On the third day of admission, the patient complained of new-onset pain and swelling in both the knee joints, and thereby a local ultrasound revealed knee joint effusion, this was tapped, and the aspirate was frank pus around 60cc from each knee joint. The knee aspirates thereby came positive for *Burkholderia pseudomallei* with sensitivity to Minocycline. Thereafter screening of the abdomen and pelvis was done in view of melioidosis to screen for abscesses in internal organs but no such thing was found on the scans. The patient was started on minocycline as per the sensitivity on the antibiogram and improvement in general condition was seen.

**Table 1: Biochemical and microbiological analysis**

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Complete blood count</td>
<td>WBCs 24.6 x 10^9/L, Hb 10.8 g%</td>
</tr>
<tr>
<td></td>
<td>Platelets 226 x 10^9/L</td>
</tr>
<tr>
<td>Kidney function test</td>
<td>Serum Creatinine 0.8 mg/dl, BUN 53mmol/L</td>
</tr>
<tr>
<td>Special Test</td>
<td>Serum Procalcitonin 100 ng/ml</td>
</tr>
<tr>
<td>Urine Culture</td>
<td>Sterile</td>
</tr>
<tr>
<td>Knee joint aspirate study</td>
<td>Cells= 2000 with 30% lymphocytes and 70% polymorphs, Proteins= 4.4 gm/dl, no coagulum Sugar = 11mg/dl, Gram Staining Gram-negative bacilli, Culture Positive for Burkholderia Pseudomallei with sensitivity to Minocycline.</td>
</tr>
<tr>
<td>Blood culture (2 sites) Day 3</td>
<td><em>Burkholderia pseudomallei</em></td>
</tr>
</tbody>
</table>

Again, on day 5 of admission, the patient developed an abscess on the right ankle joint of which pus culture was again positive for *Burkholderia Pseudomallei* with sensitivity to Imipenem and Minocycline.

As she was a referral case her previous hospital had done the biopsy for her suspected sarcoma and on day 7 of her stay in our hospital her biopsy reports arrived which were suggestive of acute suppurative and necrotizing granulomatous osteomyelitis, non-granulomatous histiocytic rich inflammation of skeletal muscle and adipose tissue, with no epithelioid granuloma or nuclear atypia. The immunohistochemistry panel was all negative with no evidence of malignancy and a core biopsy from the central core of the suspected sarcoma site was advised again by the consultant Onco-pathologist in order not to miss out on the diagnosis. Therefore, a diagnosis of sarcoma was
reconsidered and further, it was interrogated with an MRI of the left shoulder and a core biopsy of the site was performed with all due consent of patient and relatives and sent for histopathology and IHC again. The MRI (Figure 3) was now suggestive of infective etiology like Left Humerus Osteomyelitis rather than malignancy. The patient’s general condition improved, and the patient was doing fine and was vitally stable. The patient was discharged and was told to do a follow-up with histopathology and IHC reports, but all again were negative in view of malignancy.

Figure 3: MRI of the left shoulder shows a large heterogeneous STIR hyperintense signal involving the left humerus, the humeral head, neck, proximal and mid-shaft, predominantly involving the medullary cavity with cortical thinning and irregularities in the region of the head. The moderate subperiosteal extra osseous collection is seen in the proximal humerus 12 mm in thickness. Visualized deltoid muscle appears edematous with STIR hyperintense signal.

In discussion with the oncologist and oncopathologist, she was advised for CT-guided lung biopsy of secondary metastatic nodules but when she was posted for the procedure there was no evidence of such existing nodules (Figure 4), on the lung CT scan on screening as these nodules might be multiple abscesses as a sequel of melioidosis and therefore the procedure was terminated. Hence, this patient was diagnosed as a case of “Melioidosis”.

Figure 4: HRCT chest screening prior to the biopsy of lung nodules which is suggestive of normal findings
Discussion

This case report is important because it highlights a case of a clinically aggressive bone tumor viz sarcoma which turns out to be osteomyelitis. In such cases, osteomyelitis can be confused with primary bone tumors [4] but extremely rarely with sarcomas of bone. A review of the literature showed no similar cases of melioidotic osteomyelitis mimicking sarcoma. The route of spread of *Burkholderia Pseudomallei* is via aerosols or inoculation through injured skin surfaces exposed to contaminated soil or water [5]. However, modes of inoculation like hospital-acquired cases, person-to-person spread, sexual transmission, breast milk, and mother-to-child transmission are also reported to date [6-7]. There is a variation in the incubation period of this organism, which ranges from less than a day to 21 days and can extend to several months or years. Melioidosis can present as a standalone syndrome or as a component of sepsis. The disease manifests as multiple abscesses in the lungs, liver, spleen, and soft tissue, and it can cause osteomyelitis of the bone [8]. In some cases, melioidosis can remain subclinical, while in others it can present as an acute or chronic disease or even progress to fulminant fatal sepsis [9].

In our case, the diagnosis of melioidotic osteomyelitis was done against the diagnosis of sarcoma only after radiological evaluation, microbiological evidence of *Burkholderia pseudomallei* and osteomyelitis confirmed on histology. Histologically, the biopsy of this lesion revealed no malignancy which was confirmed twice when taken from the same site of suspected sarcoma with IHC reports negative both times.

Conclusion

In conclusion, osteomyelitis is a well-known source of confusion with primary neoplasms of bone. However, it is an extremely rare mimic of bone tumors like sarcoma and the presentation described in our case report appears to be unique.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his/her consent for his/her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of Interest: Nil

Financial Disclosure: None
References