Primary tuberculosis of the fibular diaphysis: A rare case report

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ABSTRACT

INTRODUCTION: Primary diaphyseal tuberculosis has very low occurrence. With no systemic signs and specific radiographic features, there exists low index of suspicion, which may delay the diagnosis of tuberculosis.

PRESENTATION OF CASE: A female aged 15 years presented with chronic leg pain and swelling for past 7 months. There was no significant history of tuberculosis present. On investigations ESR was 44 mm and positive mantoux test. Chest radiograph was normal. On x-ray (R) fibula intramedullary eccentric lytic lesion and on MRI (R) leg intramedullary lytic lesion was present suggestive of ewing's sarcoma. On histopathology epitheloid granulomas with langhans giant cells were present. Category 1 antitubercular drug regimen was started and lesion healed with alleviations of signs & symptoms.

DISCUSSION: Tuberculosis presents with typical signs and symptoms in adults compared with children in whom cystic tubercular lesions in shaft of long bones presents mostly as a single solitary intramedullary lytic lesion which corresponds with other more common differentials. This clinical and radiological heterogeneity warrants lesional biopsy and culture to determine the right diagnosis to aid in early starting of correct treatment and recovery of the patient.

CONCLUSION: With atypical presentation of diaphyseal tuberculosis in children, a high index of suspicion with unexplained pain and swelling of the bone could help to establish the diagnosis.

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1. Introduction

Pulmonary tuberculosis has more incidence than bone tuberculosis [1]. Further diaphysis involvement of long bones in skeletal tuberculosis has very low occurrence rate in pervasive areas itself (1–3%) and as well as in non-endemic areas [2]. Tuberculosis in long bones present mostly as a solitary lesion with no specific signs and symptoms [3], which leads a surgeon think of more common differentials [4] like cystic bone lesions, bone tumours, brodie’s abscess, chronic pyogenic osteomyelitis or fungal/bacterial granulomatous lesions. Also due to the rarity of incidence of disease in tubular bones [5] with no characteristic radiological sign [6] a correct diagnosis to be made is often neglected. Diagnosing such a disease requires combined surgeons knowledge of the possible differentials along with radiological investigations and assessing that with histopathological and culture sensitivity results to reach a definitive diagnosis [7]. We reported this case to emphasize the unusual occurrence of long bones tuberculosis at unusual site as in our case in fibula diaphysis area. The case has been reported in line with the Surgical Case Report (SCARE) criteria [8].

2. Presentation of case

A 15-year-old female presented to our hospital after a delay of 7 months with chief complaints of pain in right leg along with swelling for the past 7 months. Parents initially consulted non orthopaedic doctors at their local vicinity and were advised painkillers and rest but she did not get relief and then was referred to our hospital for further management. The pain was insidious in onset (parents noticed swelling 10 days after the onset of pain) progressively worsened with time but no diurnal variation was present. Pain was more on standing and walking and did not relieve on taking analgesics. There was no history of any preceding trauma, chronic cough, expectoration, respiratory complaints, evening rise of temperature, and weight loss or arthralgia. There was no significant family history of tuberculosis or contact with any tuberculosis patient present. She was afebrile and haemodynamically stable. There was a firm, diffuse, tender swelling over the lateral aspect of distal one-third shaft region of right fibula without any fixity to the overlying skin and no local rise of temperature on palpation. There was mild oedema of the overlying skin, but no discharge or pus from the lesion. The left ankle joint and knee joint were normal with no swelling, tenderness or any restriction in the range of...
movement. No lymphadenopathy was present. Rest of the systemic examination was normal. A clinical diagnosis of diaphyseal bone tumour with other possibilities including ewings sarcoma, chronic infective osteomyelitis, brodie’s abscess, cystic bone lesions were considered. Raised erythrocyte sedimentation rate of 44 mm in first hour with haemoglobin 9 g/dl was present. Tuberculin skin test was positive. Plain radiograph of the right leg showed an intracortical lytic bony lesion with adjacent sclerosis but no cortical breach in fibula shaft region (Figs. 1 & 2). MRI right leg (Figs. 3 & 4) confirmed an intramedullary diaphyseal lesion in fibula shaft, 6.2 cm away from ankle joint, with associated marked thinning of overlying cortex with focal cortical breech along anterior cortex. (suggestive of ewings sarcoma with osteosarcoma kept as another possibility). CT scan of thorax, abdomen and pelvis was performed that showed lungs were clear. The visualized skeleton was normal. Liver, pancreas and both kidneys were unremarkable. There was no free peritoneal fluid or free peritoneal air or abdominal or pelvic lymph node enlargement. Examination of the other systems was also normal. Needle biopsy was done which on histopathology showed (Fig. 5) epitheloid cell granulomas, occasional langhans giant cells and moderate mixed inflammatory infiltrate. Acid fast bacilli were negative on microscopy culture. She was put on category I antitubercular regimen, under Revised National Tuberculosis Control Programme (RNTCP) as per revised WHO guidelines [9]. Anti-tubercular therapy included 2 months of daily intensive therapy with four drugs: isoniazid (H) 10 mg/kg, rifampicin (R) 15 mg/kg, pyrazinamide (Z) 35 mg/kg and ethambutol (E) 20 mg/kg followed by maintenance therapy with drugs (HR) in the same daily doses. Currently patient is on anti-tuberculosis treatment (ATT) for past 12 months and reports alleviation of clinical symptoms with
healed lesion on plain x-ray radiograph of right fibula taken at 12 months (Fig. 6).

3. Discussion

Skeletal tuberculosis in long bones is a great mimicker of medicine [6]. Despite recent advances, tuberculosis in long bones remains a widespread problem due to its non-specific presentation with diffuse pain & swelling and very few signs of inflammation [7]. The treatment most frequently used was incision and drainage, due, in part, to early diagnosis of pyogenic osteomyelitis. With the advent of better diagnostic techniques and anti-tuberculosis treatment, saucerization & packing in the presence of the draining sinuses and saucerization & closure in cases without sinuses, is now the accepted treatment of choice in tuberculosis of the long bones of the extremities [10]. A study by Akgl T et al. mentioned an average delay of 6.6 months in the diagnosis of bony tuberculosis [11]. Published reports in literature are more on adults [12,13] and reports typical presentation in adults compared with children in whom cystic tubercular lesions in shaft of long bones [14] presents mostly as a single solitary intramedullary lytic lesion on MRI, which corresponds with other more common differentials & lead to a surgeon misdiagnosing it in early stages and start of appropriate treatment [2,5,15]. In certain cases, though imaging can help surgeon in reaching a possible diagnosis or to rule out other differentials but largely the radiographic imaging of the involved bone is not characteristic with non-specific signs such as osteopenia, sclerosis, periosteal reaction more likely to be seen [16]. Intrusive analysis is usually required irrespective of radiographic findings due to wide range of presentation and differentials [2]. Frequently diagnosis is angled towards brodie’s abscess which presents as a solitary sclerosed lytic type of lesion on radiograph and should be differentiated from tubercular osteomyelitis [17] as there are very few characteristic radiological findings and the diagnosis establishment relies mainly on histopathological and microbial culture validation [4,18]. Also, absence of response to salicylates ruled out possibility of osteoid osteoma [19]. No contrast enhancement on MRI ruled out the possibility of hemangioma [7]. Bone pain non responsive to simple analgesics made diagnosis of neoplastic or infective more likely. In a primary diaphyseal eccentric lytic lesion, as in our case, it becomes crucial to come to a conclusive diagnosis to aid in proper diagnosis, starting of correct treatment and recovery of the patient. Few case reports relating to diaphysis long bone tuberculosis have been mentioned in literature. A retrospective radiological study mentioned rare occurrence of long bones tuberculosis [20]. Richter et al. [21] mentioned importance of biopsy in diagnosing long bone tubercu-

4. Conclusion

In the absence of specific clinical features and radiographical signs, awareness of diaphyseal tuberculosis and a high index of suspicion in children with unexplained pain and swelling of the bone, could help to establish the diagnosis. Further, histopathological examination is essential for early diagnosis and accurate treatment.

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Ethical approval

Ethics approval was taken.

Consent

The patient was minor. So, we got statement of written informed consent from her parent for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of the journal on request.

Author contribution

AS was the primary surgeon in this case and conceptualized the study. AM assisted in the case and wrote the manuscript, did the proof reading, review of literature and the editing of the manuscript.

Registration of research studies

NA.

Guarantor

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Provenance and peer review

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Declaration

The necessary patient and author details have been removed from the figures before submitting.

Declaration of Competing Interest

The authors report no declarations of interest.

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