Unusual presentation of more common disease/injury

Tubercular dactylitis and multifocal osteoarticular tuberculosis- two rare cases of extrapulmonary tuberculosis

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Involvement of small bones of hand and feet leading to tuberculous dactylitis involvement is a rare presentation of extrapulmonary tuberculosis. Even rarer is the association of tuberculous dactylitis with multifocal skeletal involvement, even in countries like ours, where tuberculosis is endemic. The authors report two cases, one of tuberculous dactylitis in a 55-year-old male and another of multifocal skeletal tuberculosis in a 4-year-old male. Both the patients were effectively treated with antitubercular drugs.

BACKGROUND

Tuberculous involvement of metacarpals and phalanges is a rare presentation of extrapulmonary tuberculosis, and the occurrence of multifocal skeletal involvement is exceptional.^{1 2} The diagnosis is often delayed as many diseases of bone resemble it clinically and radiologically. These lesions respond effectively to antitubercular drugs. We hereby report two cases, one of tuberculous dactylitis

and another of multifocal skeletal tuberculosis in two non-immunocompromised patients.

CASE PRESENTATION

Case 1: A 55-year-old man presented with pain and swelling of right middle finger from 6 months. There was no history of trauma to hand. Patient gave history of fever from 2 months. There was no history of cough or weight loss. There was not any significant history.

On physical examination, his middle finger was swollen at proximal interphalangeal joint. There was pain and difficulty in moving the finger. His laboratory values showed mild anaemia (11 gm%) and raised erythrocyte sedimentation rate (60 mm at the end of 1 h by Wintrobe method). TLC was normal (6000 cells/cumm) with lymphocytosis (differential leucocyte count- P 40, L 58, E 2, M 2). ELISA for HIV was negative. Right hand radiograph showed soft tissue swelling at the proximal interphalangeal joint of the middle finger with erosion of the head of the proximal phalanx and narrowing of

Figure 1 X-ray showing proximal interphalangeal joint of the middle finger with erosion of the head of the proximal phalanx and narrowing of the joint.

Figure 2 Section showing granulomatous lesion with Langhans giant cells and epithelioid cells. (H&E x10).

Figure 3 Swelling of right proximal fourth digit.

Figure 4 Swelling on right foot.

Figure 5 X-ray showing cystic expansion of proximal phalanx of fourth digit (spina ventosa) of right hand.

the joint (figure 1). Chest radiography and abdominal ultrasound were normal. Fine needle aspiration (FNAC) of the lesion was tried which yielded only blood. Biopsy of the lesion showed exuberant granulomatous lesion with Langhans giant cells and epithelioid cells (figure 2). Ziehl–Neelson (Z–N) staining showed rare acid fast bacilli (AFB).

Case 2: A 4-year-old male child presented with complaint of swelling of right ring finger, right elbow and foot from 7 months. The patient was a known case of pulmonary tuberculosis and had completed treatment 1 year back. There was history of low-grade fever from 2 months. On examination, the patient was afebrile with height and weight below 10th percentile. Local examination revealed firm, non-tender swellings on right proximal

fourth digit, right elbow and right foot (figures 3 and 4). Local temperature was slightly raised. Overlying skin was normal. On investigations, complete blood count showed total leucocyte count (TLC) of 180000/mm³ with lymphocytosis (polymorphs 30, lymphocytes 70). There was hypochromic microcytic anaemia (haemoglobin-8.2 gm%). HIV test was negative in this patient also. X-rays of the bones showed cystic expansion of proximal phalanx of fourth digit (spina ventosa) (figure 5) along with osteolytic lesion with cystic expansion of lower end of humerus and upper end of olecranon process of ulna (figure 6) and similar lesion of cuneiform bone and first metatarsal bone of right foot (figure 7). FNAC of the lesions showed granulomatous inflammation consistent with tuberculosis (figure 8). Z–N staining showed AFB

Figure 6 X-ray showing osteolytic lesion with cystic expansion of lower end of right humerus and upper end of olecranon process of right ulna.

Figure 7 X-ray showing osteolytic lesion with cystic expansion of cuneiform bone and first metatarsal bone of right foot.

(figure 9). Chemotherapy was started in both the cases comprising of isoniazid, rifampicin, pyrazinamide and streptomycin for the first 2 months and isoniazid and rifampicin after 2 months for another 12 months.

INVESTIGATIONS

X-ray, FNAC and biopsy.

DIFFERENTIAL DIAGNOSIS

Inflammatory arthritis, pyogenic osteomyelitis, Brodie's abscess, etc.

TREATMENT

Chemotherapy was started in both the cases comprising of isoniazid, rifampicin, pyrazinamide and streptomycin

Figure 8 Fine needle aspiration of the lesion showing granulomatous inflammation (H&E x40).

for the first 2 months and isoniazid and rifampicin after 2 months for another 12 months.

OUTCOME AND FOLLOW-UP

Both the patients were effectively treated with antitubercular drugs.

DISCUSSION

Osteoarticular involvement occurs in only 1–3% of patients with extrapulmonary tuberculosis.³ 50% of these lesions are found in spine.² Involvement of the metacarpals and phalanges of the hand are infrequent.¹

The most common presentation of tuberculosis of the metacarpals and phalanges of hand is localised pain and swelling.¹ Fever and weight loss are also seen in most of the patients. Clinically it may be confused with a bone tumour or osteomyelitis.^{4 5}

This disease occurs secondary to a primary focus which can be in the respiratory, renal or alimentary tract. In about 75% cases, the primary focus is in the lungs.⁶ However, in a study by Subasi *et al*,¹ on seven cases of metacarpal and phalangeal tuberculosis, no patient had an active tubercular lesion or history of pulmonary disease as in one of our case.

Patients with disseminated skeletal tuberculosis involving hands are usually immunocompromised.⁶

Radiologic findings of osseous tuberculosis are non-specific. There may be sclerotic or osteolytic lesion.^{7 8} Spina ventosa is a rare radiologic feature with cystic expansion of the short tubular bones of hand.⁶ Other findings on plain radiograph include osteopenia, soft tissue swelling with minimal periosteal reaction, narrowing of joint space and subchondral erosions.⁹

These non-specific radiologic features may be present in inflammatory arthritis, pyogenic osteomyelitis, Brodie's abscess, etc.⁸

The diagnosis is often delayed because osseous tuberculosis is a paucibacillary lesion and synovial fluid aspirates

Figure 9 Ziehl–Neelson (Z–N) staining showing acid fast bacilli (Z–N x100).

or smears are often negative for AFB.⁸ Positive Z–N staining for AFB requires at least 10^4 AFB/ml of specimen. The gold standard for the diagnosis of osseous tuberculosis is culture of *Mycobacterium tuberculosis* from bone tissue. The advent of DNA detection via PCR increases the sensitivity of detection.^{10 11}

The treatment of osseous tuberculosis includes a 2 month initial phase of isoniazid, rifampicin, pyrazinamide and ethambutol followed by a 6–12 month regimen of isoniazid and rifampicin.¹²

To conclude, swelling and pain of bone can be a presenting sign of tuberculosis. Tuberculosis of metacarpals and phalanges of hand can be difficult to diagnose during the early stage which should be suspected in cases of long-standing pain and swelling of metacarpals and phalanges.

Learning points

- ▶ Swelling and pain of bone can be a presenting sign of tuberculosis.
- ▶ Tuberculosis of metacarpals and phalanges of hand can be difficult to diagnose during the early stage.
- ▶ Tuberculosis should be suspected in cases of long-standing pain and swelling of metacarpals and phalanges.
- ▶ Radiology, FNAC and histopathology aid in diagnosis as the lesions are paucibacillary.
- ▶ These lesions respond effectively to antitubercular drugs.

Competing interests None.

Patient consent Obtained.

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