Extensive Osteomyelitis of Humerus Following Bacille Calmette-Guérin Vaccination

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ABSTRACT
Bacille Calmette-Guérin (BCG) vaccination administered at birth can lead to certain complications which may present immediate or late. We report a rare case of BCG osteomyelitis in a nine months old immunocompetent female child with extensive destruction of middle and distal third of humerus sparing the proximal bone. Surgical debridement revealed caseous material and dead bone. Biopsy showed inflammatory tissue containing Langhan's giant cells and granulomatous inflammatory infiltrate. Culture was suggestive of BCG osteomyelitis. Anti-tubercular chemotherapy favoured betterment of her condition and the child has no signs of infection after one year.

KEY-WORDS: Bacille Calmette-Guérin; BCG Osteitis; Extensive Osteomyelitis

Introduction
Bacille Calmette-Guérin vaccination is extremely safe in immunocompetent hosts however some complications may occur.[1-3] These include abscess or ulceration at the site of vaccination and regional lymphadenitis. Rarely osteomyelitis can occur as a late complication adjacent to the site of vaccination or at a different site due to hematogenous spread.[4,5] The risk of osteomyelitis is more in immunodeficient hosts where disseminated infection can occur.[6] Our patient was immunocompetent but developed extensive osteomyelitis involving about two thirds of the humerus sparing proximal bone with separated sequestrum which was removed.

Case Report
A 9 months old female child presented to us with discharging sinus in the medial aspect of the arm. Parents complained that the child has not actively used her left upper limb for the past two months. A swelling was noticed initially that later formed a discharging sinus in the region. There was no history of trauma pertaining to the involved limb. The child was born by normal vaginal delivery to healthy parents and received BCG vaccination after birth. The child is immunized up to date under national vaccination program guidelines. There was no previous history of any illness or any family history of tuberculosis.

Examination revealed a fixed flexion deformity in the elbow of the affected side. The child did not actively use the left upper limb and there was obvious wasting of the muscles of arm and forearm. The child was irritable on handling the limb and mere handling caused pus to squirt out through the sinus. One actively discharging sinus of 0.5 mm was present in the medial aspect of middle third of arm. Passive movement of elbow from 30 degrees of fixed flexion to 90 degrees was possible. Shoulder range of motion was normal. There was no distal neurovascular deficit.

Mantoux test was performed showing induration of 12 mm. ESR and CRP were elevated. Patient had normal humoral and cellular immune function. Radiograph revealed extensive destruction in diaphyseal and distal metaphyseal region of left humerus along with sequestrum separated from normal bone and lying within a lucent lesion (Figure 1). Clinical picture of this patient comes under the Foucard's criteria[4] for BCG osteomyelitis satisfying four of the five points proposed by him, (1) Patient had a BCG vaccination; (2) It is less than four years since vaccination; (3) Lacking contact with tuberculosis; (4) Histopathological picture indicates tuberculosis. An only criterion that is not satisfied
is the radiological picture which is more extensive than those described in the literature.

Diagnosis of BCG osteomyelitis was confirmed with tissue culture from biopsy specimen.

Anti-tubercular therapy was given consisting of rifampicin (10 mg/kg body weight per day), isoniazid (10 mg/kg body weight per day) and ethambutol (15 mg/kg body weight per day). Injection streptomycin (30 mg/kg body weight per day intramuscularly) was given as long as she was an inpatient. Pyrazinamide was not used because it is generally considered ineffective for the treatment of BCG osteomyelitis.[5-7] Plaster of Paris support was given and radiograph at one month was satisfactory (Figure 2). The therapy was continued for 8 months and there were no signs of recurrence. Sequestrectomy, curettage and appropriate chemotherapy was the key to child's recovery. One year follow up of the child reveals no signs of infection and an improved range of motion of the elbow.

**Discussion**

BCG vaccine replaces the potentially dangerous primary infection due to M. tuberculosis with an innocuous primary infection due to bacilli of Calmette and Guerin which activates the host cell mediated immunity.[8] This vaccination protects children mainly from tuberculous meningitis and disseminated tuberculosis.[9] It is of more use to those living in countries where tuberculosis is still common. When a person is exposed to potential M. tuberculosis, it will be only a reinfection for which the body had already produced immunity.[8]

Although certain complications of BCG vaccination exist, hematogenous spread causing osteomyelitis is rare. BCG ositis involving distant sites like the lower extremity, sternum and ribs are also reported in the literature.[3] In most reports, patients had a normal chest X-ray and did not have any systemic complications but typical radiological and histopathological findings.[4,9,10] In our case, the condition was extensive with gross destruction of bone and formation of sequestrum. It is mandatory to assess the immunological status in such patients.

Diagnosis of BCG osteomyelitis should always be kept in mind and can be confirmed with multiplex PCR. Many studies have shown growth of a strain
that is resistant to pyrazinamide and hence it is not included in their chemotherapy regime.\[5,7\]

Timely interference and appropriate chemotherapy is the key to faster recovery of the patient. Though a standard chemotherapy regime for BCG osteomyelitis is not available, prolonged treatment with antitubercular agents for 8-10 months is followed in most of the studies and the rate of recurrence is very less.\[5,7,11\]

**Conclusion**

In conclusion, BCG osteomyelitis is not common. Even though extensive osteomyelitis of humerus as seen in our case can occur, it responds well to surgical debridement and timely chemotherapy. It can well be treated without any complications in an immunocompetent patient.

**References**


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