The Bone Scan Pattern in Disseminated BCGitis

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ABSTRACT

Despite the long history of the worldwide use of Bacillus Calmette-Guerin (BCG) vaccine, a wide spectrum of adverse reactions has been observed in a small proportion of immunized infants; the most severe complication, disseminated BCGitis, is often fatal but exceedingly rare and is considered to result from host immunodeficiency.

At present, CT scan, ultrasound, X-ray and bone marrow aspirations are the investigations used to diagnose this disease. This is the case report of a 6 months old female infant with disseminated BCGitis. This paper aims to highlight the advantages of the use of nuclear medicine imaging and pattern of bone scan for diagnosis of disseminated BCGitis.

Key words: BCG vaccine, Disseminated BCGitis, Bone scan

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INTRODUCTION

The original BCG strain of mycobacterium bovis was derived from multiple passages of wild type M.bovis (1). BCG vaccine is administered worldwide to prevent tuberculosis and is considered to have an excellent safety profile. A live bacterial vaccine, BCG should not be administered to persons with immune system impairment (2,3). In such individuals BCG may disseminate from the injection site to multiple organs, usually with fatal consequences (4,5,6).There is a few cases of report disseminated BCG specially reported from Iran, and Canada(7,8). However, there has been no report of bone scan for diagnosis and evaluation of extent of the disease.

This is the case report of a 6 months old female infant, who was diagnosed with disseminated BCGitis. The aim of this study is to present the

important role that nuclear medicine bone scintigraphy in diagnosis of this disease.

CASE PRESENTATION

A 6 months old female infant, presented to our hospital due to irritability and respiratory distress. There was a 3 week history of irritability and fever with respiratory difficulty, for which she had been treated by antibiotics. She later developed an oral lesion along with diarrhea and vomiting and had to be admitted to hospital with suspicion of pneumonia and meningitis.

The only notable past medical history was hyper bilirubinemia at day 4 of birth, which was treated by phototherapy. Parents were first cousins. The patient's brother, 9 months old, had died the previous year with suspicion of pneumonia. Initial examination revealed fine crackles in the right lung and tongue thrush. A course of penicillin and acetaminophen was prescribed.

X-ray of the skull showed a single lesion in the right side of the sagittal suture, which was suggestive of vascular compression. Abdominal ultrasound showed a heterogeneous hypoechoic round lesion in the anterior lateral aspect of the right hepatic lobe. This was thought to be suggestive of mesenchymal hematoma. Enlargement of the spleen with normal homogeneous echo texture was also seen. Abdominal CT scan demonstrated mild hepatosplenomegaly.

Chest X-ray showed moderate to severe pleural effusion in the right side. There were multiple lytic lesions noticed in multiple ribs. For whole body bone evaluation, bone scan was performed. Following IV injection of 300 MBq Tc-99m Methylen diphosphonate (MDP) whole body bone scan was performed which revealed multiple active bony lesions in the skull, ribs, pelvic bones, both entire both femorea, tibiae, and entire upper limbs (Fig 1).The scan pattern was highly suggestive of disseminated bony inflammatory process (osteomyelitis) and with respect to the patients age, possibility of an immune deficiency disorders was suggested.

Then for better evaluation X-Ray from pelvis, femur and forearms was performed showing multiple lytic lesions (Fig 2). Bone biopsy was performed and with respect to scintigraphic findings, culture of the specimen was requested which was positive for acid fast bacilli. Diagnosis of disseminated BCGitis was made. A course of anti-tuberculosis medication was commenced.

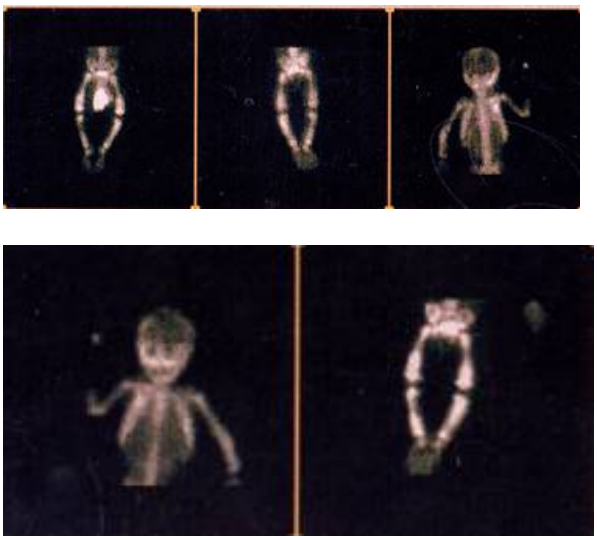

Fig 1- Whole body bone scan of the patient

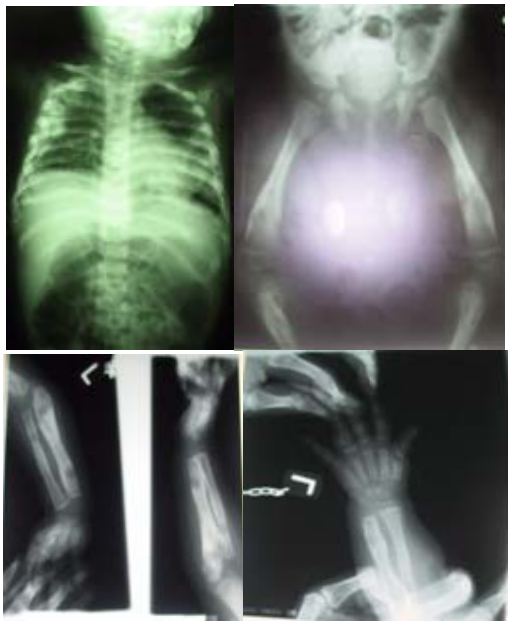

Fig 2- Radiographies of the patient.

Despite treatment, the patient's condition deteriorated. She developed hepatosplenomegaly with jaundice, bilateral temporal edema, 3 part aphthous oral lesions, maculopapular facial rash and papules on the abdomen. Full blood count revealed leukopenia (91% neutrophils) and anemia with increased ESR and CRP levels. The patient expired 6 weeks after admission.

DISCUSSION

Disseminated BCGitis is a rare, yet fatal side effect of BCG vaccination in infants. Both local and systemic BCG vaccine-associated complications continue to occur. It usually presents as generalized lymphadenopathy, skin rash and hepatosplenomegaly (9). Disseminated BCGitis rises as a result of impaired immune system of the host. Complications of BCG vaccination can be severe and life threatening in infants with impairment of immune system. Immunodeficiency is classified into primary and secondary. The most common secondary immunodeficiency leading to disseminated BCGitis is HIV infection. However, considering parental consanguinity in this case and the brother's death at a similar age following an illness with similar clinical picture, there is a high probability that she had a primary immunodeficiency.

Severe combined immunodeficiency (SCID) with autosomal recessive pattern of inheritance, interferon γ receptor deficiency (IFNγR1 and IFNRγ2 deficiencies) and interleukin 12 (IL-12) deficiency are some of the primary causes of immunodeficiency that predispose the individual to disseminated BCGitis (6, 9-10).

The clinical presentation of disseminated BCGitis is not pathognomonic and the physician can be misled. For example, the occurrence of systemic manifestations, such as skin rash and hepatosplenomegaly, can closely mimic those of haematological malignancies (11). Bone marrow aspiration and, in the case of lymph node involvement, fine needle aspiration of lymph nodes involved are the best methods for definite diagnosis of disseminated BCGitis (6, 11). Currently, there are no reports of the use of nuclear medicine imaging in the diagnosis and evaluation of the extent of bony involvement in disseminated BCGitis. The bone scan is a non-invasive method with low radiation burden which can be easily performed leading to early diagnosis of this disease.

In conclusion, disseminated BCGitis is a rare dangerous adverse effect of BCG vaccination in infant with immunodeficiency. At present, CT scan, ultrasound, X-ray and bone marrow aspirations are the investigations used to diagnose this disease. However, considering the availability and non-invasive nature of bone scintigraphy and its leading pattern in the presence of disseminated BCGitis, it is recommended in the diagnosis of BCG osteitis/ osteomyelitis.

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