Short Communication

Retroperitoneal Abscess due to Disseminated Bacille Calmette-Guérin Infection

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SUMMARY: Disseminated mycobacterial infection after Bacille Calmette-Guérin (BCG) vaccination is a rare disorder, usually presenting with fever, weight loss, anemia, lymphadenopathy and hepatosplenomegaly. We report a case of disseminated BCG infection in a 28-month-old girl with prolonged fever and abdominal mass due to retroperitoneal abscess. Appropriate therapy resulted in a good response. This is the second reported case of retroperitoneal abscess complicating BCG vaccination.

In Iran’s expanded immunization program, Bacille Calmette-Guérin (BCG) vaccine is given at birth. It has relatively high protective efficacy against meningeval and miliary tuberculosis in children, but immunocompromised individuals, especially congenital phagocytic or cell-mediated immunodeficient infants and HIV sufferers, are at risk of disseminated BCGitis syndrome (1). With disseminated infection, the organs most commonly affected are the lymph nodes and bone marrow. Involvement of the lung, liver, spleen skin and bones are relatively common (1). The most commonly reported findings are fever, lymphadenopathy, hepatosplenomegaly and weight loss (1). Abscess formation is rarely reported in organs distant from the injection site (2,3). This is the second reported case of disseminated BCG infection with retroperitoneal abscess.

A 28-month-old girl was admitted in Nemazee Hospital, affiliated to Shiraz University of Medical Sciences in southern Iran, with the chief complain of high-grade fever and sweating especially at night for 2 weeks. She had been immunized with the BCG Pasteur strain 1173P2 at birth and developed a normal superficial ulcer at the site of injection in the extensor site of the right upper arm 1 month later, which healed with a scar within 2 months. Suppurative lymphadenitis in the right axial area had appeared at the age of 1 year, and was excised surgically, with acid-fast bacilli seen in the pus obtained in the operation room. One month after surgery, another suppurative lymphadenitis was observed over the right lateral cervical area; it spontaneously healed with a scar 3 months after fistulization.

The pertinent findings in the abdominal palpation were limited to a splenomegaly and an ill-defined mass in the right upper quadrant area which could not clinically be differentiated from hepatomegaly. A 2 × 3 cm erythematous nontender lymph node, ruptured without forceful palpation, was found in the right inguinal area. The bacteriologic study of the discharge was negative for aerobic culture and Gram’s staining. Excisional biopsy revealed caseating granulomatous inflammation with positive acid-fast bacilli.

Laboratory studies revealed leukocyte of 17,200/mm3 (81% polymorphonuclears, 16% lymphocytes, 3% monocytes), hemoglobin of 6.7 gr/dl, platelet of 421,000/μl, an erythrocyte sedimentation rate of 38 mm/h, C-reactive protein of 96 mg/dl and serum protein of 6.2 g/l. The liver function test, Wright test, Widal test, and indirect immunofluorescence (IFA) test for kala azar, and urinalysis were within normal limits. Urine and blood cultures and malaria smears were negative. Tuberculin skin test was non-reactive. Bone marrow aspiration was negative from the point of malignancy, lipid storage disease and leishmaniasis. Chest x-ray was normal. A whole-body bone Tc-99m scan revealed increased uptake at the L3-L4 vertebrae, suggestive of an abnormal pathological process. In abdominal ultrasonography, the liver size was normal. An enlarged spleen (about 103 mm) with a normal echo pattern and a confined fluid-filled space in the right lower quadrant were noticed. In the abdominal CT scan, a large retroperitoneal abscess (11 × 5 cm) extending from the upper part of the right kidney to the pelvis but having no attachment to the right kidney was detected (Fig. 1).

Fig. 1. Abdominal CT-scan with contrast of a case of retroperitoneal abscess due to disseminated BCG infection. 1A reveals a collection of fluid with a thick outer wall in some aspects in the right retroperitoneal space. An enlarged spleen with homogenous density also is evident. 1B shows the extension of the collection to the pelvis medial to the iliac crest. Marked displacement of the right ureter is seen.
exploration of the abdomen confirmed the presence of a retroperitoneal collection with non-malodorous pus without any adjacent lymphadenopathy. Gram’s stain, aerobic and anaerobic cultures of pus obtained in the operation room were negative, but numerous acid-fast bacilli were detected in the smear. The culture was positive for *Mycobacterium* spp. The presence of the BCG strain of *Mycobacterium bovis* was confirmed by polymerase chain reaction (PCR) administrating a primer common to all known BCG strains (4).

Our attempt to detect any clue to immunological problems such as failure to thrive, side effects of other vaccines, immunosuppressive administration, suggestive family history or suggestive past history of recurrent, severe or unusual infection, was negative. The levels of serum IgG, IgM and IgA, CH50 and the nitrotetrazolium blue dye test were also normal. We were not able to perform the advanced immunological tests that show individuals’ predisposition to severe mycobacterial infections such as interferon (IFN)-γ level, IFN-γ and interleukin (IL)-12 receptors.

The patient, who appeared to have disseminated BCG infection, was treated with isoniazid and rifampin. She showed good response to the therapy in a 2-month follow-up examination.

Disseminated BCG infection is defined as the presence of infection at two or more anatomical sites beyond the region of vaccination and is mostly accompanied by a systemic syndrome compatible with mycobacterial disease (1). Our 28-month-old patient presented with persistent fever, inguinal lymphadenitis, splenomegaly, retroperitoneal abscess and vertebrae osteomyelitis. Acid-fast staining and pathological features of the excised inguinal lymph node pointed to mycobacterial disease. Acid-fast bacilli were detected in the material of the abscess, and BCG strain of *M. bovis* infection was confirmed by PCR.

In the literature, lymph nodes have been reported as a common site of involvement in disseminated BCG infection (1,6). Hence, the suppurative inguinal lymphadenitis in our patient led us to suspect the disease. Lymph node involvement in patients suspected of disseminated BCG infection may play a key role in the diagnosis.

In many cases of disseminated BCG infection, a perforate suppurative axillary lymphadenitis at the same site of the BCG injection is an early finding of the disease (5,6). The present case had a history of draining axillary BCG lymphadenitis 16 months prior to the admission, which might be considered as the onset of the disease. Once suppurative axillary lymphadenitis is presented ipsilateral to the BCG injection site, disseminated BCG infection must be considered and requires more investigation.

Although most cases of disseminated BCGitis syndrome are in patients under 2 years of age, the disease can manifest many years after vaccination based on the immunity status of the host (1,7). This case developed into full-blown disease at 28 months of age.

Although retroperitoneal abscess is a rare presentation of disseminated BCGitis disease, and to date only one other case of the disease has been reported in the literature (2), the disease should be considered as a potential cause in cases of abdominal abscess.

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**REFERENCES**