Leishmaniasis with Femoral Bone Involvement

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ABSTRACT

Visceral leishmaniasis is mostly subclinical, but it can become symptomatic and take acute, subacute or chronic forms. Its incubation period varies from weeks to months and can even be as long as years. A vast variety of organ involvement may be there. We report a case of osseous leishmaniasis presenting with only a long standing mild knee pain without any laboratory or organ abnormality and showed a pure bone involvement on X-ray and CT scan. Later on, it developed into a discharging sinus and the discharge showed Leishman bodies which was reconfirmed the rough bite. As the patient had reactions with amphotericin B, systemic glucantime was given which led to clinical improvement and smaller radiological lesion.


INTRODUCTION

Leishmaniasis is a parasitic infection that is transmitted by the bite of female Phlebotomine sandflies. Mammals including human beings are reservoirs of leishmaniasis. Leishmaniasis occurs as three main forms: cutaneous, mucosal and visceral. Cutaneous form is the most common form of leishmaniasis. Visceral leishmaniasis (VL) is the most severe and life-threatening form of leishmaniasis with mortality rate of 10%,¹ ²

According to the World Health Organization (WHO) report, more than 90% of cutaneous leishmaniasis occurs in Afghanistan, Algeria, Brazil, Colombia, the Islamic Republic of Iran, Peru, Saudi Arabia and the Syrian Arab Republic.² More than 90% of visceral leishmaniasis occurs in Bangladesh, Brazil, Ethiopia, India, Nepal and Sudan.² The incubation period of visceral leishmaniasis varies from weeks to months and can even be as long as years. VL affects the vital organs including liver, spleen, and bone marrow and causes different symptoms like fever, weight loss, splenomegaly, hepatomegaly, and anaemia.³

Here, we report a case of visceral leishmaniasis that presented only with a long period of mild knee pain, due to osseous involvement only.

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CASE REPORT

A 23-year man came with a dull pain around his left knee that had been there for more than 8 years. Since then, many different evaluations had been performed without any conclusive result and he had been treated symptomatically.

On physical examination, there was no movement restriction (passive or active) and the examination was normal. On knee radiography, there was a mild lytic lesion at the distal femur in anteroposterior and lateral X-rays of the knee (Figure 1). CT scan was done showing a lytic lesion in the condyles and distal part of the femur, accompanied by scalloping and sclerosis around it in a remarkable geographical pattern and cortical thickening without destruction and periostalization (Figures 2a, 2b). Two months later, a fistula with discharge was appeared in the lateral part of the same knee. A specimen was taken from discharge and sent for pathological study. In histological study, a high number of Leishman bodies was reported.

The patient was admitted for biopsy. During the procedure, a completely destructive cystic lesion covered with a thin membrane was observed. A biopsy specimen was taken by curettage of the cyst membrane which confirmed the diagnosis of Leishman body. Both results were rechecked by a reference laboratory and confirmed. Also, the result of discharge culture for Leishman body was positive.

Blood biochemistry was normal. Involvement of other organs, including spleen and liver, was ruled out by abdominal sonography and CT scan.

After the diagnosis was confirmed, treatment was started with amphotericin-B but adverse reactions (hypotension and dyspnea) limited the treatment. Due to this, treatment was switched to systemic glucantime. After 6 months of treatment, the pain improved and the lesion size reduced in CT scan.
DISCUSSION

Leishmaniasis is endemic in 99 countries, and 72 of these are developing countries. There are about 350 million patients with leishmaniasis throughout the world and every year 200,000 - 400,000 new cases are reported. The classic cause of VL in Asia is Leishmania donovani and its major locations are lowland trait region of southern Nepal, Bangladesh, Bihar province, and surrounding areas in India, China and Pakistan.

VL can have an extensive spectrum of symptoms that vary from being asymptomatic to Kala-Azar, with presentations like fever, weight loss, and involvement of bone marrow that can lead to pancytopenia and hyper gammaglobulinemia.

Sub-clinical form of VL presents with non-specific mild clinical symptoms lasting for a long period with fluctuating course. In this patient, there was no abnormality in the biochemical tests or symptoms and signs except for a chronic non-specific mild knee pain that was not accompanied by any other symptoms initially, and without any outstanding change. The only positive point in the patient history was living in an endemic area for leishmaniasis (Mashhad).

Diagnosis of VL is difficult in the early disease phase before development of the classic triad of fever, splenomegaly and pancytopenia. This may cause delay in diagnosis. In addition, diagnosis of parasite in the spleen, liver, and bone marrow needs intervention, which further increases the complexity of diagnosis. Also, VL may mimic symptoms of connective tissue disease and misdiagnosed as connective tissue disorder. Due to these difficulties, diagnosis of VL in this patient was delayed.

VL is treated with antiparasitic pentavalent antimonial agents, liposomal amphotericin B, oral miltefosine, pentamidine and other agents. Liposomal amphotericin-B is the first choice and the only one that is licensed for VL in the United States. Pentavalent antimonial agents, such as paromomycin sulfate, pentamidine, and glucantime are second-line therapies for VL.

Due to allergic reactions to the first choice agent in this patient, the second-line agent was started. The pain improved and the follow-up CT scan showed a smaller lesion after 6 months of treatment.

Leishmaniasis should be considered in differential diagnosis in patient with non-specific chronic symptoms, belonging to endemic areas.

REFERENCES