

Case Report**Visceral leishmaniasis in a patient with cutaneous lesions, negative Leishman-Donovan bodies and immunological test: A case report**

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Abstract

We report a case of visceral leishmaniasis (VL) in a patient from Shush in Iran with cutaneous lesions, negative for Leishman-Donovan bodies, enlarged spleen filled by leishmania protozoa and negative immunological test. The patient was a 26-year-old male, who attended hospital with fever and two deep purulent lesions on the distal part of his left leg. On physical examination, the patient had splenomegaly. Laboratory results were as follows: pancytopenia, positive C-reactive protein (CRP), elevated erythrocyte sedimentation rate (ESR) and lactate dehydrogenase (LDH) levels. The necessary treatment was administered to the patient. Biopsy of lesion for Leishman-Donovan body was negative. In addition, indirect fluorescent antibody (IFA) screening for leishmaniasis was negative. Diagnostic splenectomy was performed which pathological exploration showed a bulk of leishmania protozoa in patient's spleen. Twenty days later, this patient expired.

KEYWORDS: Visceral Leishmaniasis, Cutaneous Lesions, Leishman-Donovan.

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Leishmaniasis is a cluster of diseases caused by protozoa in the genus leishmania¹ that are transmitted by the bite of infected female phlebotomine sandfly.² Leishmaniasis afflicts the populations of 88 countries around the world.³ This parasitic disease has a broad spectrum of clinical manifestations which are attributable to the leishmania species.⁴ The disease presents in four different forms: visceral leishmaniasis (VL) or "kala-azar", cutaneous leishmaniasis (CL), mucocutaneous leishmaniasis (MCL) and diffuse cutaneous leishmaniasis (DCL). Visceral leishmaniasis may present with an acute, subacute or chronic course, but most patients remain completely asymptomatic.² Because of the increase in international travel and immigrant populations, the numbers of cases of leishmaniasis is probably to increase.⁵ There are an estimated 500000 new cases of visceral leishmaniasis per

year in the world.⁶ Therefore, it is important that clinicians are able to diagnose the unusual presentations of leishmaniasis.⁵

We describe a case of visceral leishmaniasis associated with cutaneous lesions, negative Leishman-Donovan bodies and immunological test in a patient from Shush, a county in Khuzestan Province in Iran.

Case Report

A 26-year-old male from the Shush (a county in Khuzestan Province in Iran), referred to Al-zahra Hospital, Isfahan, Iran, with fever and two deep purulent lesions for past one month. The lesions (about 8 × 4 cm in diameter) were surrounded by erythematous and edematous borders and central necrosis. Lesions were evident on the distal part of the left leg, fixed-size at the beginning and accompanied by foul-smelling yellow discharge. The gross feature of

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lesions was similar with CL (Figur 1). Considering research ethics, the past history and the present clinical data were recorded in a data sheet. The patient was a known case of Down syndrome. On his recent hospitalization, he was diagnosed as suffering from myelodysplastic syndrome (MDS) by a bone marrow aspirate (BMA) and bone marrow biopsy (BMB), following a graduate pancytopenia. The patient was treated with cefixime, itraconazol and ciprofloxacin, without any improvement in clinical conditions.

In addition, he was on treatment with phenytoin compound due to the epilepsy from childhood. Physical examination showed that the patient had splenomegaly. The results of his blood tests are depicted in table1. Blood culture, antinuclear antibody (ANA), antineutrophil cytoplasmic antibody (ANCA), coombs' test, chest X-Ray (CXR) and echocardiography (ECG) did not show any abnormality. Smear and culture of wound discharge were negative

for fungus and mycobacterium. Abdomino-pelvic ultrasonography showed a spleen span about 145 mm. Doppler ultrasound of lower limbs was normal and no sign of deep venous thrombosis (DVT) was seen. Massive soft tissue swelling was seen by the lower limb sonography, and soft tissue computed tomography scan impressed cellulites and myositis. On hematological consultation, BMA and BMB were recommended for the patient but no abnormal finding was detected. He received Granulocyte colony-stimulating factor (G-CSF) but no improvement in blood count was seen. Treatment was started with meropenem (1g q8h), vancomycin (1g q12h), cefepime (2g q8h), metronidazol (500mg q6h), cotrimoxazole (2 tablets q8h), rifampin (300mg q12h) and amphotericin B (50 mg/day). Amphotericin B was discontinued due to the severe side effects such as fever, chills, headache, nausea, vomiting and phlebitis at the site of the infusion.



Figure 1. Deep purulent lesions on distal part of the left leg

Table 1. Laboratory evaluations for the patient

Test	Result	Normal ranges
WBC count (cells/ μ l)	1100	4500-10000
PMN (%)	50	55-75
Hemoglobin (g/dl)	6.7	14-18
Platelet count (cells/ μ l)	25000	150000-450000
Reticulocyte count (%)	1%	< 2%
CRP (mg/L)	positive	negative
ESR (mm/hr)	50	20
LDH (U/L)	3770	5- 450

WBC: with blood cell, PMN: polymorphonuclear leukocytes, CRP: C-reactive protein, ESR: erythrocyte sedimentation rate, LDH: lactate dehydrogenase

Because of the gross appearance of the lesions which was similar to CL, skin biopsy was taken from lesions which was negative for Leishman-Donovan bodies in three times recheck. In addition, indirect fluorescent antibody (IFA) screening for leishmaniasis was negative. For definite diagnosis, diagnostic splenectomy was performed, which pathological exploration showed a bulk of leishmania protozoa in patient's spleen. Twenty days later this patient expired.

Discussion

This is a case report of VL with cutaneous lesions in Iran which was negative for Leishman-Donovan bodies and immunological test. Leishmania can affect the skin, viscera, or mucocutaneous areas, varying according to the geographic region and the host response.⁵ Leishmaniasis directly affects hundreds of thousands people per year, with millions of individuals at risk worldwide.⁷ Leishmaniasis can manifest with various coetaneous manifestations, based on the genetic differences within the host or the parasites.⁸ The diagnosis of CL is made on the basis of a typical lesion, a history of exposure and demonstration of the parasite on the lesion.⁹ This infective skin disease manifests as localized, often self-healing single lesions, intermediate forms which frequently produce mucosal lesions.¹⁰ Many of infected patients often experience a delay in diagnosis and inappropriate treatment.¹¹

Our case was an interesting one with some cutaneous lesions, grossly similar to CL, with-

out any Leishman body inside. In addition, it seems strange that an enlarged spleen full of leishmania protozoa but without positive IFA (indicator of VL) was found. In this new case, we cannot rule in/out CL, due to dermatological presentation and absence of Leishman-Donovan bodies. Pourahmad et al. reported a case of human immunodeficiency virus / visceral leishmaniasis (HIV/VL) co-infection associated with cutaneous lesions in a patient from Jahrom, in Iran. They demonstrated that it is better to evaluate the diagnosis of visceral leishmaniasis in patients who present with cutaneous leishmaniasis and HIV infection.² In our case, regardless of the spleen filled with Leishmania protozoa, VL could be ruled out due to the negative IFA. One interpretation for this case may be the accompanying MDS. Since serological abnormalities like hyper- or hypogammaglobulinemia is reported to be about 18-65% in the cases afflicted by MDS,¹² it may mask the proper immunological response to the leishmania parasites. Level of anti-leishmania antibody in immunocompromised patients is 50 times lower than those with normal immune system.¹³ The inconsistent finding was the absence of Leishman-Donovan bodies in the wound smears, that is not interpretable with MDS. The more reasonable suggestion that we can make is that the patient may be afflicted with VL after the CL. But the spared bone marrow in a patient with a spleen full of Leishman-Donovan bodies is interesting, if the diagnosis is VL.

Leishmaniasis is an endemic disease of Iran that unfortunately, there is no definite treat-

ment for this disease.¹⁴ The epidemiological trends of concomitant CL and VL cases have changed today, which needs further attempt for better diagnostic/therapeutic approaches. Our case demonstrated that negative smears

for Leishman-Donovan bodies in cutaneous lesions similar to CL, and negative IFA in suspected VL should not draw us away from the diagnosis of leishmaniasis in the suspected cases.

Conflict of Interests

Authors have no conflict of interests.

Authors' Contributions

FKh, AEN, MB participated in diagnosis and treatment of the patient. FA Participated in writing and editing the final version of the report. All authors have read and approved the content of the manuscript.

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