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Case report Open Access

Cutaneous leishmaniasis in patient with polyarteritis nodosa vasculitis: A Case report

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Cutaneous Leishmaniasis is the most prevalent type of leishmaniasis. Iran is among the first six countries in the world with the highest annual incidence of cutaneous Leishmaniasis. Non-caseating granulomatous inflammatory cutaneous reaction is the most common pattern of its skin lesions. The current article presents the case of a 36-year-old man with a history of polyarteritis nodosa vasculitis with extensive caseating granulomatous lesions who was diagnosed with cutaneous leishmaniasis. Caseating granulomatous can present in cutaneous Leishmaniasis; thus, it is recommended to consider leishmaniasis in the diagnosis of this histological pattern, especially in autoimmune patients before starting immunosuppressive treatment.
Keywords: Caseating granulomatous; Cutaneous leishmaniasis;Autoimmune disease; Case report

Introduction

Leishmaniasis is a common zoonotic group of diseases with general manifestations appearing in three main types: cutaneous leishmaniasis (CL) (most prevalent), mucosal leishmaniasis (ML), and visceral leishmaniasis (VL), the most severe type potentially resulting in fatality [1, 2].

The global prevalence of leishmaniasis is currently estimated to be at least 14 million in 82 countries. The annual infection rate has been declared to be two million, while at least 350 million others are considered still at risk. In 2017, the number of new cutaneous leishmaniasis cases was estimated to be at least 1.5 million each year [3].

Concerning the disease burden and distribution, about 88 countries worldwide are endemic for cutaneous leishmaniasis, and 90% of patients are found in Afghanistan, Algeria, Brazil, Pakistan, Peru, Saudi Arabia, and Syria. In 2013, the Center of Disease Control in the Ministry of Health, Iran, announced the incidences of cutaneous leishmaniasis in different provinces. According to this report, the highest incidence rates occurred in Ilam (102.6 per 1000), Fars (86.7 per 1000), and Khorasan Razavi provinces (63.9 per 1000), and the lowest incidence (0.01) was seen in Gilan [3].

Iran is among the first six countries in the world with the highest annual incidence of cutaneous leishmaniasis. It seems the incidence of CL is underestimated in all countries [2]. The spectra of clinical manifestation of CL ranges from small cutaneous nodules to gross mucosal tissue destruction

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Therefore, it isconsidered as a group of diseases in the tropics and neo-tropics [4]. The prevalent clinical cutaneous signs and symptoms of CL involve papules leading to ulcers and symptoms such as fever, headache, weight loss, and loss of appetite. These are not especial manifestations in most tropical and subtropical regions that are endemic for various infectious diseases such as tuberculosis, human immunodeficiency virus (HIV), malaria, and typhoid, which often present with similar cutaneous symptoms and may complicate and delay the diagnosis of CL [5].

Cutaneous species of leishmania infect the inflammatory monocyte-derived macrophages and dendritic cells of the host. The consequences of infection depend on the host immune system, nutritional status, infectious parasites, and concomitant infections. Leishmaniasis manifestations might present as indicative of autoimmune disease. Accordingly, the immune status of these patients should be evaluated for immune response, resistance, and susceptibility to leishmania [1].

Other differential diagnoses with clinical spectra similar to leishmaniasis are leprosy, skin cancers, and cutaneous mycoses which are common in leishmaniasis-endemic areas [4]. In 2016, one study on CL reported an unexpectedly high number of caseating granulomas (equal to the Ridley group C), which was not in line with other research [6].

Herein, we aimed to present a case of cutaneous leishmaniasis with caseating skin granulomatous lesions in a patient with polyarteritis nodosa (PAN).

Case presentation

The patient was a 36-year-old man with a history of PAN vasculitis from the age of 12 years old. Because of recurrent infection from that time, cytotoxic drug treatment had not been completed; however, chronic use of corticosteroids had led to exogenous Cushing's. The other complications of PAN vasculitis during the patient's life included muscle atrophy, being bedridden, and bedsores (but not in this admission) as well as wrist drop and foot drop due to neuropathy. He also had a history of CMV (cytomegalovirus) and pneumocystis jirovecii lung infection about 15 months prior to the current admission, which led to this long hospitalization. In his drug history, valproate sodium (for mood disorder), gabapentin, prednisolone 10 mg/qd, and cotrimoxazole 480 mg/qd for pneumocystis pneumonia (PCP) prophylaxis had been prescribed according to the patient's condition. In this admission his main symptoms were chronic extensive skin lesions on the right forearm, proximal of the right thigh, and some patches on his shin (Figure1). These lesions were erythematous with yellow exudates and painless crust. The patient complained of dysuria, agitation, anxiety, and sleep disorder. Clinical examination revealed cutaneous striae due to Cushing's. His laboratory results showed neutrophilia (84.2%) and a slight elevation in sodium level of serum (146 mg/dl). In the patient's urine analysis and culture results, pyuria was detected, and the urine culture identified Pseudomonas aeruginosa which is susceptible to ciprofloxacin; hence, treatment was initiated. Tests for Hepatitis B and C were negative.

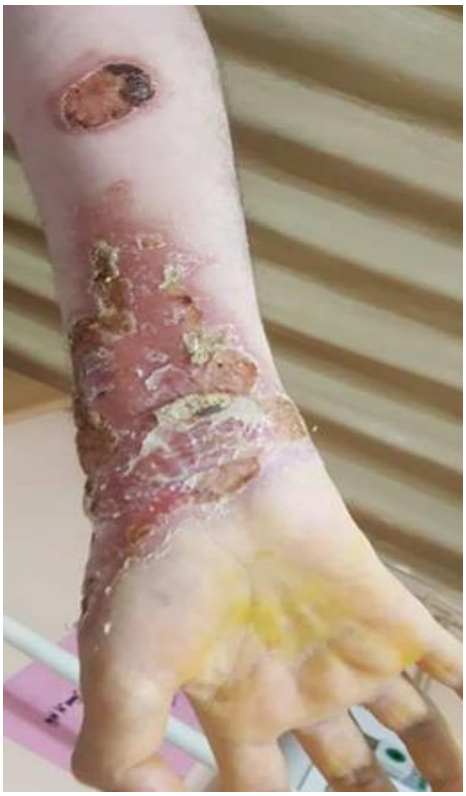

Figure 1. Forearm cutaneous lesion

In clinical assessment, the patient was found to suffer from diabetes mellitus. Ultrasound imaging revealed fatty liver (grade 2) and splenomegaly. Kidney assessment showed kidneys of normal size with

with increased echogenicity of the parenchyma, and heterogeneous reduction of renal parenchymal thickness was detected. The patient had recurrent urethral stricture that required dilatation with orology intervention. Color Doppler ultrasonography was performed to evaluate the patient's renal arteries; the resistance index (RI) on the right side was 0.62 and on the left side was 0.6, and acceleration times on both sides were normal. All renal arteries and veins showed normal blood flow on both sides. It is noteworthy that evidence of renal parenchymal infarction, hydronephrosis, or renal artery stricture was observed.

The patient's cardiac condition was assessed by echocardiography, which indicated that left ventricular hypertrophy (LVH) could be caused by hypertension or cardiomyopathy, and dilated ascending aorta (4.3 cm) was also detected. Furthermore, the patient had a normal left ventricular ejection fraction (LVEF) (equal to 55%) and history of recent PSVT.

Based on these clinical and paraclinical evaluations, initial treatment was performed to stabilize the patient's condition, including controlling the patient's diabetes by prescribing insulin and psychiatric counseling to treat the patient's agitation, anxiety, anxiety, and sleep disorder. The psychiatrist's diagnosis was major depression, and he prescribed citalopram and perphenazine. Unfortunately, after taking these medicines, the patient experienced arrhythmia seemingly related to the medications.

For the most important step, i.e. diagnosis and treatment of the patient's main complaint, a dermatologist was consulted. The first differential diagnosis was pyoderma gangrenosum, atypical mycobacterium, fungal infection, and atypical pemphigus. Therefore, skin biopsies of the lesions were taken. Smears for fungi and mycobacterium in polymerase chain reaction (PCR) were negative. Biopsy revealed inflammation with granuloma and necrosis in the center of the granuloma and caseation (Figures 2 and 3).

At this point, tuberculosis (TB) was the first possible diagnosis. Skin lesion samples presented ulceration and granuloma, suspicious for leishmaniasis. The differential diagnosis for leishmaniasis skin lesions included fungal infection, atypical mycobacterium (marinum), inflammatory disease as pyogenic granuloma, malignant neoplasm and metastasis, psoriasis, Wegner, stasis and traumatic ulcers, lethal midline granulomatosis, nasopharyngeal carcinoma, and lymphoma.

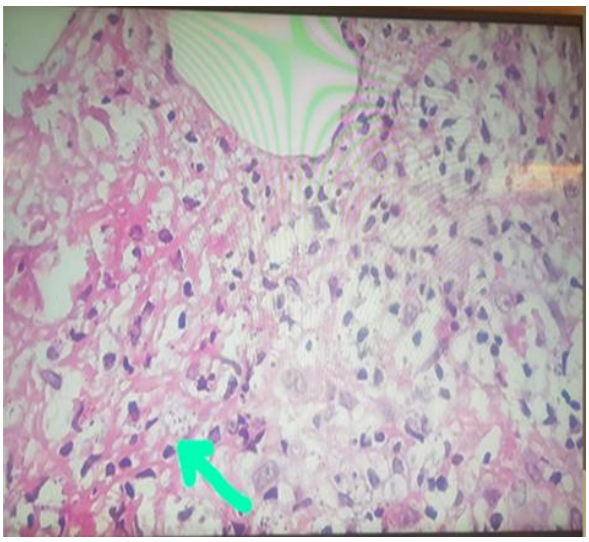

Figure 2. Microscopic view of Leishman's body

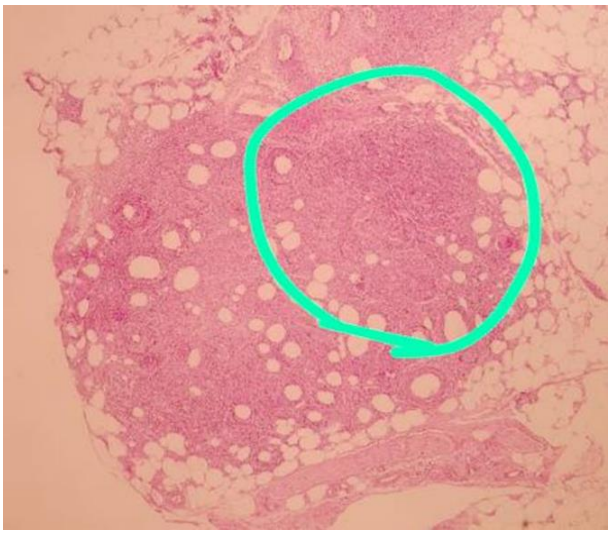

Figure 3. Caseating granuloma in a skin lesion's specimen

Caseating granulomas usually manifest in TB and rarely in cases of cutaneous leishmaniasis [7]. In the present case, the most probable diagnosis for pyoderma gangrenosum was cutaneous leishmaniasis (CL). Accordingly, treatment was started with one dose of glucantime (20 mg/kg/) intramuscularly daily for 28 days. Sadly, the patient died of Covid-19 (coronavirus) disease during treatment.

Discussion

This paper reported a case of cutaneous leishmaniasis with extensive cutaneous involvement. The histopathology pattern of the lesions was caseating granulomatosis, which is rarely seen in CL. The patient was a 36-year-old man with a history of PAN vasculitis from 12 years of age. He complained of skin lesions on his right forearm, proximal of the right thigh, and some patches on his shin. These lesions were erythematous with yellow exudates and crust without pain. According to his pathology evaluation report, the first diagnosis was TB; with more precise assessment, however, cutaneous leishmaniasis was diagnosed. Despite the administration of glucantime, the patient became infected by Covid-19 which caused his death. In a multiregional cohort study, Aoun et al. (2014) evaluated 195 cases of CL and found a notable 18.2% of all CL cases with caseating granulomas in TB-endemic regions [7]. The similarity of the geographical region of Iran, being endemic for TB, and the host immune response could explain the identical results. In 2016, Hayani et al. studied CL epidemiology and features after war in Syria. He reported nine unusual clinical expressions of CL. The cases included a dermatitis atopic with mild xerosis, an acnetic papules and nodules with the diagnosis of "familial type" CL, a chickenpox, sarcoidosis, adnexal tumors of the skin, mainly syringomas and/or trichoepitheliomas with dermatopathology result of non-caseating granulomas with amastigotes and confirmation of CL, ulcerated lesion on the forehead, Kimura's disease, ulcerated plaque in the lower hairline of a patient's beard, and herpes. The common significant characteristic in all these cases was non-caseating granulomas in cutaneous leishmaniasis, despite atypical presentations [8]. The important point of this study is the incidence of non-caseating granuloma in CL, even in unusual manifestations of it, which is particular to the case reported herein. The current article is a presentation of extensive granulomatous skin lesions in a PAN patient in which the diagnosis of caseating granulomatosis in the histopathology of cutaneous leishmaniasis is unique.

Conclusions

The important aspects of the present case are the rarity of this pathology pattern in cutaneous leishmaniasis, the differential diagnosis of TB, and other similar histopathology in these types of skin lesions. Our searches revealed only rare similar reported cases. In summary, it is suggested that cutaneous leishmaniasis in occurrence of caseating granuloma histological pattern be ruled out, especially in autoimmune patients, before starting immunosuppressive treatment.

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Conflict of interest statement

The authors have declared that they have no conflicts of interest.

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