Cutaneous leishmaniasis associated with human immuno deficiency virus infection

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Abstract

Cutaneous leishmaniasis(CL) which was relatively unknown, has become one of the leading infectious skin diseases in Sri Lanka over last two decades. It is caused by dermotrophic *Leishmania donovani* species in Sri Lanka. There area number of case reports of CL associated with Human Immunodeficiency Virus(HIV) infection in literature but not reported yet in Sri Lanka. A male patient presenting with multiple lesions of CL was later found to be infected with HIV. He was treated with courses of intramuscular sodium stibogluconate injections on two separate occasions. Initially he responded well but developed recurrences on both occasions. Patient is awaiting further treatment for second recurrence. Managing the case has become a challenge due to patient's poor compliance to treatment, and non-availability of more convenient treatment regime.

Introduction

Cutaneous leishmaniasis is a vector borne infectious disease transmitted by sandfly caused by different species of leishmania parasites in different geographic settings of world. It is caused by dermotrophic *Leishmania donovani* species in Sri Lanka[1]. It remained a rare disease with few imported cases up to 1990s. There was rapid increase in number of cases since 2000. It has become endemic in most parts of country with more than 3000 new cases being reported in 2020[2]. Number of cases reported from Kegalle district has started to increase since 2016 and local transmission is well established in some areas of the district. Sri Lanka still has a low prevalence of HIV infection with a total of about 4000 confirmed cases by the end of first quarter of 2021[3].

Though there were several reports and studies of CL associated with HIV infection in world literature, this association has not been reported in Sri Lanka. Atypical presentation and poor response to standard treatment was noted in many of these cases.

Case

A 54 year old male patient, who was a resident of Kegalle presented to skin clinic in September 2019 with 6 slowly enlarging erythematous nodules and plaques on his left forearm for about 3-6 months duration. They were painless, non-pruritic, non-scaly lesions. The patient was a security officer working in private sector, and denied overseas travel or travel to areas where CL is highly prevalent. Since the incidence of CL had a rising trend in Kegalle district, a clinical diagnosis was made even though the appearance was atypical. The diagnosis was confirmed with a skin biopsy and intra-lesion sodium stibogluconate injection was administered. The patient defaulted treatment after the first injection.

The patient came back to clinic on February 2020 and on this visit he had new asymptomatic skin nodules with central ulcer on his left ankle region and multiple small papules on lower leg (Figure 1A). The lesions previously injected had developed crusting on top of the plaques (Figure 1C). Histology of the biopsy taken from this new lesion has confirmed the diagnosis of CL. Slit skin smear and culture which had been performed on 5 skin lesions at the Parasitology Department, University of Colombo had positive results as well.

On the same visit patient complained of nail changes of recent onset and on examination he had onychomycosis of 4 fingernails (Figure 1B). Since he had atypical presentation and associated paronychia, blood was sent for HIV screening which became positive.

The patient's HIV status was confirmed by Western blot method and he had low CD4 count (96/mm^3) with high viral load at the time of diagnosis. According to history it was concluded that he had acquired HIV infection several years back. The patient was commenced on highly active antiretroviral therapy treatment (tenofovir, emtricitabine, efavirence combined tablet + co-trimoxazole 960mg daily) for HIV infection which he had continued without interruption up to now.

Considering the patients clinical picture a decision was taken to investigate for possible visceral leishmaniasis and treat him with a course of systemic sodium stibogluconate. Further assessment of patient did not reveal any clinical features of visceral leishmaniasis. His blood counts were normal and

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bone marrow aspiration and biopsy was negative for leishmaniasis. He did not have hepatomegaly or splenomegaly in ultrasound scan.

There was COVID-19 pandemic related lockdown of the country during this period and commencement of treatment was delayed as a result. He had developed 4 more small nodules of CL on his back at the time of starting treatment. Patient was commenced on a course of intramuscular sodium stibogluconate 8 ml daily for 21 days in May 2020. Due to high serum amylase the daily dose had to be reduced to 4ml and the total duration increased to 28 days. All of the skin lesions got completely resolved by the end of treatment.

Patient was lost to follow up for 5 months. He came back to the clinic on November 2020 with features of recurrence of CL at some of the previous sites on his back and left forearm (Figure 2A, B). In addition lesions showed multiple small satellite papules adjacent to initial lesions (Figure 2B). His CD 4 count had increased to 379/mm^3 and viral load was zero by November 2020. He did not have any clinical or investigation findings compatible with visceral leishmaniasis.

He was started on a second course of intramuscular sodium stibogluconate 5 to 6 ml daily with frequent monitoring of serum amylase level. This time few of the lesions remained unresolved at the end of 28 days of treatment. Patient refused continuing further injections at that point citing socio-economic issues, thus liquid nitrogen cryotherapy was applied for the unresolved lesions.

He was lost to follow up for several months and came back with a second recurrence. He had developed new lesion on his left ear (Figure 3A), in addition to the increasing number of satellite papules around the old lesions (Figure 3B). The patient is awaiting further treatment for second recurrence.

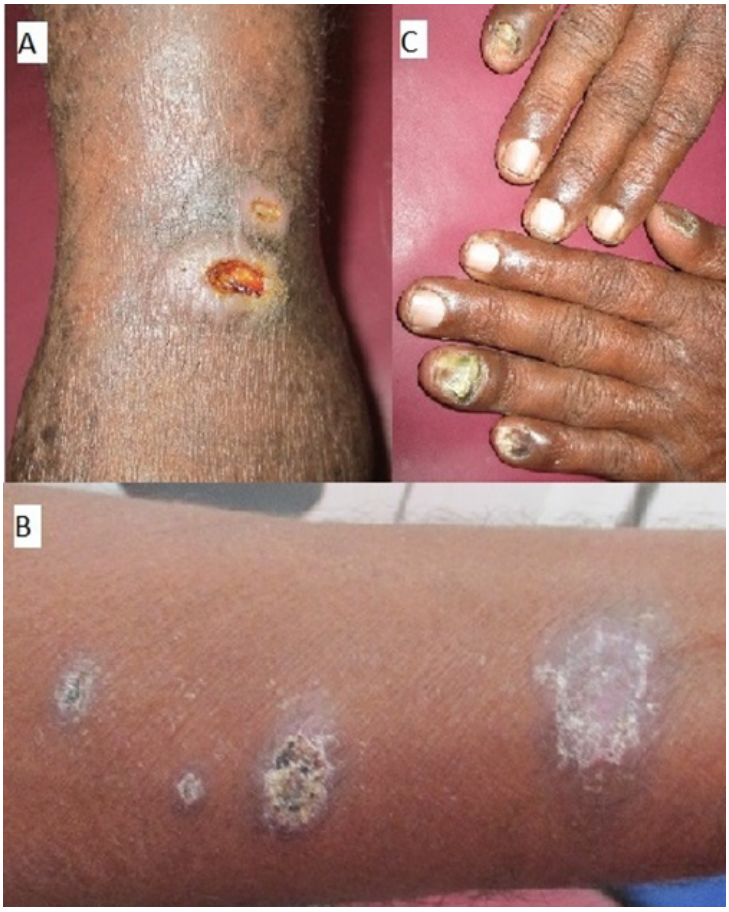

Figure 1. Features at the time of diagnosis of HIV.

A – Nodulo-ulcerative lesions at left ankle
B – Crusted plaques on left arm
C – Onychomycosis of fingernails

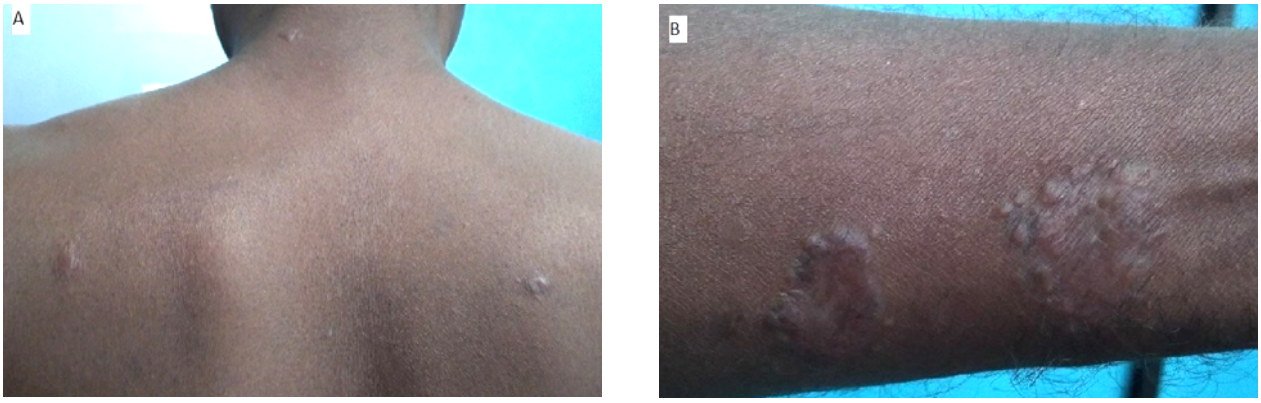

Figure 2. *First recurrence with developing satellite papules.*

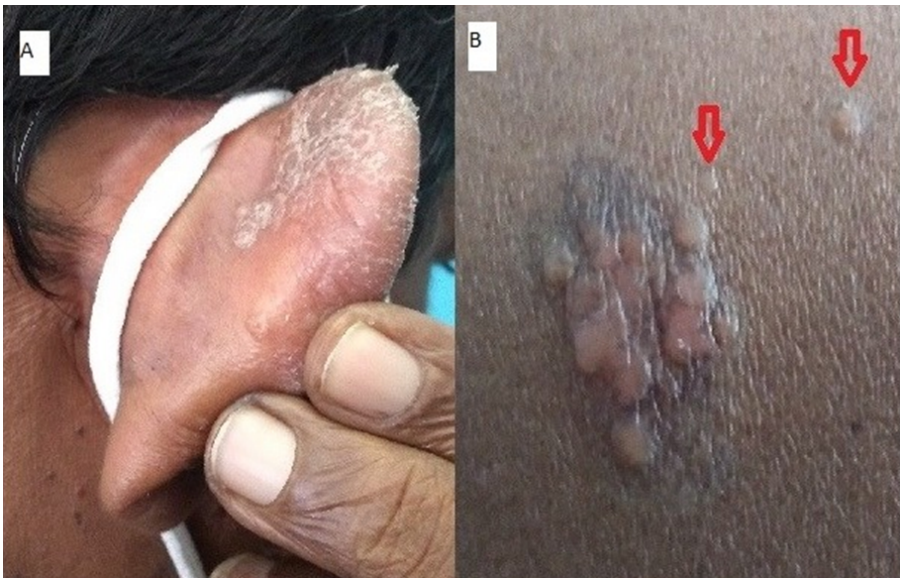

Figure 3. *Second recurrence, A – Infiltration of ear lobe, B – New satellite lesions on back of trunk (arrows).*

Discussion

There are several case reports of CL associated with HIV infection in literature caused by different species of Leishmania[4,5,8,9,10]. In our patient species identification was not performed. Since the patient had no history of foreign travel it is justifiable to assume that this was caused by dermotrophic *L donovani* species which is the causative organism in Sri Lanka.

Atypical presentation including diffuse CL, multiple lesions reported in association with HIV infection[4,5,6,7,8,9,10]. Dissemination of lesions was attributed to defect in host cellular immunity in HIV associated patients[7,10]. Our patient had both atypical lesions and few typical nodules / nodulo-ulcerative lesions. He had dissemination of disease to distant sites of skin, but the number of lesions were low and his presentation can not be labeled as a case of diffuse CL. He had not developed features of visceral leishmaniasis which is in keeping with dermotrophic nature of *L donovani* species in Sri Lanka.

He was treated twice with intramuscular sodium stibogluconate but his disease had recurred on both occasions. Satellite lesions developing adjacent to old lesions was a notable feature observed in our patient. He had developed new lesions at distant sites after the initial diagnosis, many of those on areas covered by cloth. This favours cutaneous dissemination of

disease rather than new lesions due to re-inoculation through sandfly bites.

Poor response to standard treatment and recurrence disease are features noted on many of the previously reported cases[4,5,8,9,10]. It was same with our patient. His daily SSG dose had to be reduced due to rising serum amylase. In order to compensate for lower dose it was decided to extend his treatment duration. However it was not possible to extend due to patient's socio-economic issues and it may have attributed to poor response to treatment.

Miltefosine had been used successfully to treat both visceral and disseminated cutaneous leishmaniasis in immunocompromized patients with HIV[11,12]. Miltefosine is an oral medication, thus frequent hospital visits are not necessary making it more convenient to patients. This highlights the importance of having this drug to treat patients facing a similar scenario.

It was the atypical presentation associated with onychomycosis which lead to the diagnosis of HIV infection in our patient. Since HIV screening facilities are freely available at most of the government hospitals in Sri Lanka it would be useful if CL patients with atypical presentation and poor treatment response can be screened for HIV infection in future.

Acknowledgement

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