Cutaneous Leishmaniasis in Natives of Central Region of Nepal

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ABSTRACT

We report two cases of cutaneous leishmaniasis in natives of central region of Nepal. The first patient in our series, an adult female, presented with a small nodule on the philtrum of upper lip and the second case, a male child, presented with two crusted plaques on forehead. The final diagnosis was based on histopathological findings; however, species characterization was not possible because of its unavailability in the country. These patients responded well to the treatment with Miltefosine (First case) and Fluconazole (second case). Moreover, these cases sparks a question about the origin of diseases in this region and calls for further research in future to find out the cause and prevalence of this disease in Nepal. This case report also emphasizes to consider cutaneous leishmaniasis as differential diagnosis for granulomatous presentations in our context.

KEY WORDS

Cutaneous leishmaniasis, Fluconazole, Miltefosine

INTRODUCTION

Visceral Leishmaniasis is common in Nepal, however, cutaneous leishmaniasis (CL) remains rare, though they have been reported in migrant Nepalese workers.[1-3] There are recent case series in CL in natives of western part of Nepal, which borders Himanchal pradesh in India where the disease is endemic and thus explaining its origin.[4,5] We report two cases (one adult and one child) of CL in natives of central region (Kathmandu and Kavre) of Nepal without any history of visit out of this area. The cases were initially misdiagnosed as lupus vulgaris because of rare presentation of cutaneous leishmaniasis in our scenario. The case highlights the presence of CL in natives of Nepal contrary to previous position of leishmaniasis in visceral form present only in terai region and opens an area of research to carry out epidemiological studies in our scenario.

CASE REPORTS

Case 1

A 40 year old female, housewife by occupation, from Kathmandu district presented (February 2015) with asymptomatic small nodule at the philtrum with swelling of the central part of upper lip of one year duration (fig. 1a). It started as small sized elevated skin lesion and gradually increased to a size of one and half centimeters square.

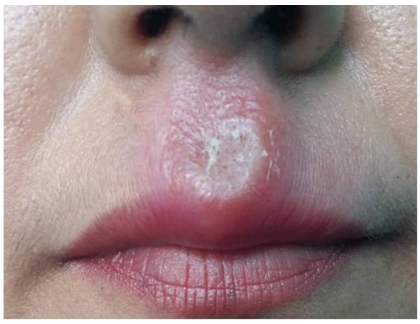

Figure 1a. **Scaly erythematous nodule on Philtrum**

There was no history of fever, weight loss, insect bite or trauma in that area. There was no history of any symptoms suggestive of major illnesses. There was no history of international visit. On cutaneous examination, there was a well demarcated non-tender single erythematous nodule of 1.5X1.5 cm with surface scaling on philtrum without regional lymphadenopathy.

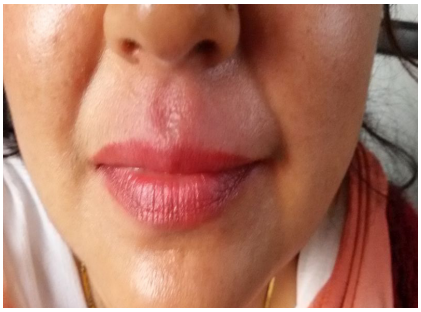

Figure 1b. **Healed lesion after a course of Miltefosine**

Complete blood count, liver function, renal function tests, and chest x ray was normal. Mantoux test was positive. Acid fast stain of slit skin smear and sputum were negative. Fungal elements were also not seen. Initial biopsy made us to think lupus vulgaris, a common presentation, based on presence of epithelioid granuloma and thus anti-tubercular treatment (ATT) was started. Since there was no response to ATT after 6 months, diagnosis was revised and a repeat biopsy was done which revealed multiple granulomas composed of lymphocytes, histiocytes with amastigotes. Leishmania Donovani (LD) bodies were seen in Giemsa stain. Patient was treated with oral Miltefosine 50 mg three times a day for 28 days. The lesions started regressing after 28 days of therapy and there was complete resolution of symptoms (fig. 1b). Consent was taken from patient for publication of the report.

Case 2

A 7 year old male child from Kavre district, presented (May 2018) with two coalescing erythematous plaques with yellowish crust over the left side of forehead since 4 months (fig. 2a). It started as papules gradually increasing in size to become a crusted erythematous plaque. There was no significant history of major illnesses in past and symptoms suggestive of systemic diseases. On examination, there was a single crusted non- tender erythematous crusted ulcerated nodule of 1.5X1.5 cm size on left forehead. Complete blood count, liver function test, chest X ray was normal. Mantoux

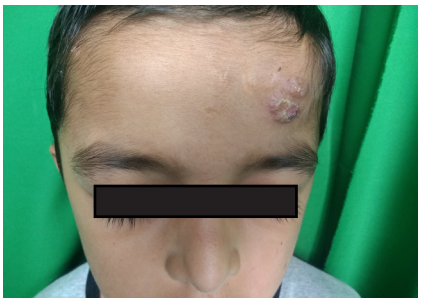

Figure 2a. **Erythematous plaque with crusts on left forehead**

test was negative. Biopsy showed multiple epitheloid granulomas along with scattered multinucleated giant cells and dense chronic inflammation in dermis. Numerous LD bodies like structures within the histiocytes were seen with final diagnosis of cutaneous leishmaniasis (fig. 2b). He was started on syrup fluconazole at 7.5 mg/kg per day for four weeks after doing his liver function test, following which he showed rapid resolution of lesion (fig. 2b). Consent was taken from patient for publication of the report.

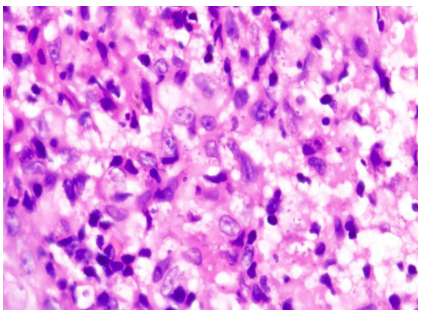

Figure 2b. **High power (H&E) with LD bodies**

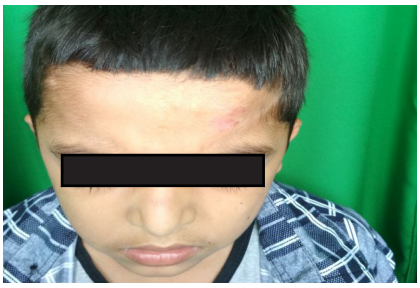

Figure 3. **Healed lesion after a course of Fluconazole**

DISCUSSION

Visceral leishmaniasis is endemic in Nepal and their cutaneous presentation in form of Post kala-azar dermal leishmaniasis (PKDL) has been reported in past.[6] This case series from central hilly region along with recent report of 33 cases in six years on cutaneous leishmaniasis from mid- western Nepal gives us clear picture of rising trend of this disease in natives of hilly and mountainous region of our country and proves that cutaneous leishmaniasis is no longer uncommon in our scenario. Cases of CL reported in past by us and few others reflected presence of CL in Nepal because of travel and immigration patterns.[3] This case reports further supports our previous observation of presence of CL in indigineous population of Nepal from the central region. Moreover, literatures remind us that

Leishmaniasis donovani which is supposed to be prevalent in our context as a cause of visceral leishmaniasis and PKDL is not supposed to cause CL in general. This notion was supported by R kumar who reported Leishmaniasis Major as a cause in one of the native Nepalese patient of CL.[7] Because of the shift of trends of visceral leishmaniasis towards hilly and mountainous region as mentioned in annual report from Department of health services ,Nepal and presence of CL in natives in these regions, it is high time to carry out epidemiological research work for vector and species characterization in this region of Nepal.

Leishmaniasis is a disease caused by intracellular protozoan parasite (genus Leishmania). Infection is transmitted by the bite of the sand fly. With the exception of Australia, the Pacific Islands, and Antarctica the parasites have been identified throughout the large portions of the world.[8]

Leishmaniasis is simply classified based on clinical categories as Cutaneous, mucocutaneous, visceral and viscerotropic leishmaniasis. Cutaneous forms are further sub- divided into localized, diffuse, recidivans, and post-kala-azar dermal leishmaniasis.[8]

A 'classical' lesion starts as a papule or nodule at the site of inoculation; it grows slowly, taking at least 1 week to reach its final size. A crust develops centrally, which may fall away, exposing an ulcer up to 5 cm in diameter with a raised edge and variable surrounding indurations, which heals gradually over months or years, leaving a depressed scar with altered pigmentation. Satellite nodules at the edge of the lesion are common.

Diagnosis of cutaneous leishmaniasis is by history and clinical examination, which can be confirmed by demonstration of amastigotes in Giemsa stained smears from infected skin by direct microscopy, intracellular amastigotes in the dermis of H & E (Haematoxylin and Eosin) stained sections from biopsy specimen, presence of leishmanial granulomas in the dermis in H and E specimens, growth of promastigotes in Nicolle-Novy- macNeal (NNN) culure medium from lesional specimens and demonstration of Leishmanial DNA by PCR. The scarcity of amastigotes in the lesion can easily lead to delayed or incorrect diagnosis. Because of constraints of laboratory diagnosis, we have been largely dependent on clinical features and histopathological demonstration of amastigotes for diagnosis of cutaneous leishmaniasis.

Pharmacologic therapies includes sodium stibogluconate,liposomal amphotericin B, Oral miltefosine intramuscular pentamidine, ketoconazole, fluconazole, itraconazole, allopurinol and dapsone.[8]

Because of unavailability of sodium stibogluconate, we used miltefosine in adult case and fluconazole in child, with favourable response in 4 weeks time.

Because of non-endemicity of CL in Nepal and heterogenous presentation, it can easily be missed clinically and can be treated as some other forms of granulomas like cutaneous tuberculosis or leprosy which was similar in one of our case. These cases are the first reports of CL in natives of central region of Nepal. The presentation of disease in child could be indicating endemic CL in our setting. It also highlights the importance of considering cutaneous leishmaniasis in patients presenting with granulomatous lesions and that Miltefosine and oral fluconazole can be given as an effective alternative regimen in places where pentavalent antimony are not available. Furthermore, this article calls for carrying out nationwide research to redefine the status of cutaneous leishmaniasis and characterize the species, and the concerned authority to make the drugs easily available for treatment of this condition.

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