Cutaneous leishmaniasis: an emerging infection in a non-endemic area and a brief update

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Abstract
We report here the emergence of a new focus of cutaneous leishmaniasis (CL) due to *Leishmania tropica* (*L. tropica*) in the Ajmer city of Rajasthan, India, a previously non-endemic area. Between January-February 2006, 13 new indigenously acquired cases of CL were diagnosed among the patients attending the Skin and STD department, JLN Hospital, Ajmer. The diagnosis was based on clinical presentation, demonstration of amastigotes (LT bodies) in Giemsa stained smear of the lesion and response to intraläsional / local anti-leishmanial drug therapy. In addition, culture of the promastigote forms of *L. tropica* from the lesion was successfully attempted in four of the smear negatives cases. By retrospective analysis, 23 new indigenous cases of CL have been diagnosed in the same setting during the period January 2004 - December 2005, based on clinical and therapeutic response alone. There was no clear-cut history of sandfly bite and travel outside the district or state to endemic area in any of the cases. However, all of them came from a common residential area (famous dargah of Ajmer) and the peak incidence was seen in January, four months after the famous Urs fair of Ajmer, the location was urban and the lesions were characteristic of *L. tropica*. Therefore, the disease is suspected to be anthropogenic. These features are suggestive of a common mode of transmission, source and/or vector signalling introduction of this infection into a non-endemic area.

Keywords: Ajmer, cutaneous leishmaniasis, emergence, *L. tropica*, non-endemic region

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Cutaneous leishmaniasis (CL), first descriptions of which can be traced back to the ninth century (Balkh sore), remains a major world health problem in the 21st century. The Leishmaniases are caused by the protozoa *Leishmania*, which is transmitted by the bite of an infected female sandfly of the genus Phlebotomus (Old world CL) or Lutzomyia (New world CL). The result of infection can vary from acute or chronic skin ulcer, recurrent or diffuse CL to erosive mucosal disease with progressive destruction of the nasopharynx and severe facial disfigurement, to a life-threatening systemic infection with hepatosplenomegaly, though very rare. The resulting syndrome depends upon a complex interaction between a specific species of *Leishmania* and the genetic and immunological status of the host. Several species of *Leishmania* are involved including *L. major*, *L. tropica* and *L. aethiopica* (together known as *L. tropica* complex) on the old world and species of *L. braziliensis* and *L. mexicana* in the new world.

[1][3] New world species do not occur in India.
There are about 1.5 million new cases of CL each year, of which over 90% occur in seven countries, namely: Afghanistan, Algeria, Brazil, Iran, Peru, Saudi Arabia and Syria. However, CL often occurs in specific pockets. In India, indigenous cases of CL are confined to the dry northwestern half of the Indo-Gangetic plain, including dry areas bordering Pakistan from Amritsar to Gujarat. To the east, cases have been reported from as far as Delhi and Varanasi in U.P. [4] Surveys carried out during 1973 in whole of erstwhile endemic areas revealed the presence of sporadic cases in Fazilka (Punjab), Rajasthan canal zone at Hanumangarh (Sriganganagar district), Jodhpur city and rural and urban areas of Bikaner district in Rajasthan. [4] Ajmer district of Rajasthan was hitherto not a known endemic focus for CL. In this communication, we report emergence of CL in the Ajmer city of Rajasthan for the first time.

~ Materials and Methods

During the period of 2004-2005, 23 new clinically suspected, indigenous cases of CL were diagnosed in patients attending the Skin and STD clinic, based on retrospective analysis. It is interesting to note that more than ninety percent of such cases were residents of a common focal (famous Dargah) area of urban Ajmer and were mostly Sindhis or Muslims. But, the depth of the problem came to light with the occurrence of a cluster of 13 new cases within a span of two months in January-February, 2006. The diagnosis of CL was based on clinical presentation, demonstration of amastigotes in Giemsa stained smear of the lesions and response to intralesional and local anti leishmanial drug therapy.

Slit skin or aspirate from lesion was taken in 11 out of 13 patients (two of the cases refused to co-operate in the procedure and were spared as one of them was a child aged six years and the other was a young female with lesion over the nasal cartilage) and stained with Giemsa for demonstration of LT bodies. In addition, the aspirate from the lesion in smear negative or doubtful cases was inoculated onto Schneider's drosophila medium (Hi-Media) as well as Toebies biphasic medium containing rabbit defibrinated blood, prepared in-house. For each patient, detailed information regarding age, sex, caste, residence, occupation and history of travel outside the district or state especially to endemic areas for CL, history of bite by sandfly and clinical features of the presenting lesion, was taken. Since, detailed information regarding the aforementioned aspects was not available by retrospective analysis, CL cases (n=23) diagnosed during 2004-2005 was not included in the final results.

All the diagnosed adult cases were treated with 1-5 mL of sodium stibogluconate intralesionally by injection till blanching of the lesion, [5] once weekly for minimum three weeks or till clinical cure, whichever was earlier. Larger dose was used for multiple lesions. In case of children, local ketoconazole (2%) was administered twice daily for 10 days. The patients were followed-up for cure.

~ Results

Clinical details of the cases and important results of the study are shown in the [Table - 1]. In all cases, the duration of disease at the time of presentation was one to four months. The predominant type of lesion was red, non-ulcerative indurated plaque of size 1-2 cm. The lesion was pruritic in only four of the cases. Ten patients had single lesion including all the five children. Face was afflicted in seven patients and the lesions were confined to cheeks, cartilage of the nose or the skin above the lip. In three of the patients the lesions were either on the dorsum of hand or on the extensor aspect of forearm closer to the wrist. In the remaining three patients, the lesions were multiple. Mucosal involvement was not seen in any of the patients. There was no satellite lesion or lymphadenopathy either.

There was no clear-cut history of bite by sandfly. None of them gave a history of travel outside the district or state in the preceding two years. Interestingly, six of them gave history of visit and stay in their house by nearby relatives or guests from other countries (Pakistan, Israel, Bangladesh) during Urs fair or otherwise.

Tissue smears for LT. bodies were positive in seven out of 11 (63.6%) patients. LT. bodies were present abundantly, both intracellularly within skin macrophages and extracellularly [Figure - 1]. In one of the smears, the diagnosis of LT. bodies was doubtful as they were sparse and of bizarre shape and size. The remaining three smears failed to show presence of LT. bodies. However, all the four (3 smear negative and 1 smear doubtful) cases yielded promastigotes in culture in both Schneider's and Toebies medium, thus confirming our clinical diagnosis.

Weekly intralesional injection of sodium stibogluconate was successful within one month in curing the lesion in seven out of eight adult cases treated. One of the adult cases was lost to follow-up and hence not included in the results. Local ketoconazole (2%) twice daily was seen to be effective in curing the lesion in all the five children within 10 days. Mean duration of recovery was 20 days. Patients with multiple lesions took longer to cure.

~ Discussion

Ours is a tertiary care divisional hospital catering to a population of over 21 lakhs in and around Ajmer district. Ajmer is not a hitherto known endemic area for CL and this disease was rarely encountered in patients attending our Skin and STD clinics till 2003. Since 2004, sporadic cases of CL have been witnessed clinically. By retrospective analysis, 23 new indigenous acquired cases of clinically diagnosed CL were identified from 2004-2005. In the month of Jan-Feb 2006, 13 new indigenous cases of CL were diagnosed in the same setting. This alarming increase in incidence of new cases prompted us to thoroughly evaluate the disease problem in our area.

Mode of infection and disease transmission

Ajmer city is a world renowned Muslim pilgrimage center because of Khwaja Moinuddin Chishty’s Gharib Nawaz (dargah), where people of various religions from all over the country and abroad visit throughout the year. The Urs fair of Ajmer is also famous and Muslim pilgrims (Zaireen) from all over the world come to attend this fair in huge numbers. In 2005, Urs fair held in the month of August at the Dargah was attended by approximately two lakhs Zaireen. The recent emergence of CL in the Dargah area seems to be due to introduction of the disease by infected tourist(s). This is akin to the study of RC Sharma et al. [6] who...
diseases as well. The need for medical microbiologists to enable prompt diagnosis of such diseases cannot be overemphasized.

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herald the onset of an epidemic, if not diagnosed and controlled in time. With the emergence of more and more new cases of CL

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factors, had similar clinical presentation and incubation period, responded equally well to anti-leishmanial therapy and had

Pentavalent antimony compounds remain the drug of choice for treatment of CL. All of our seven cases treated with

However, the important link in the transmission of the infection (i.e., the sandfly vector) remains unidentified due to lack of a

history of fly bite. Sandflies are the only known vectors for CL and speculating that this was the vector in all our cases, we can only hypothesize the natural evolution of the disease. The infection is transmitted from the human source (infected resident of the dargah area or an infected pilgrim from an endemic area) or animal reservoir (infected dogs or rodents inhabiting the dargah area, though a remote possibility) to the susceptible by the bite of the vector sandfly. Alternatively, the infection might be transmitted by direct contact with the lesion of an infected person or mechanically through bites by various flies. [10] This is supported by the fact that in India anthropogenic transmission with no intermediate host has been observed. [11] Demonstration of parasite in direct smears remains the easiest and one of the specific methods of diagnosis. The smears were positive for LT bodies in 63.2% cases, which is comparable to the study of Kubba et al. [12] and Sharma et al. [6] Culture yielded promastigotes abundantly in all the four cases.

The fact that all the cases came from a common focal area (Dargah), were mostly Muslims or Sindhis, had no documented risk factors, had similar clinical presentation and incubation period, responded equally well to anti-leishmanial drug(s) is yet to be seen.

The outpatient visit at our Skin and STD clinic suggests that there are probably many more smouldering cases of CL, which may herald the onset of an epidemic, if not diagnosed and controlled in time. With the emergence of more and more new cases of CL and the possibility of an epidemic, we have alerted the local public health authorities to investigate the epidemiology of the disease at Ajmer. As the disease is suspected to be anthropogenic, control of further spread may be achieved with comparative ease.

This, we suppose is a classical example of how a disease can be introduced and spread rapidly among virgin population in an otherwise non-endemic area to become ultimately indigenous to that area, by means of travel, communication and clustering. It also highlights the need for people of the medical fraternity to be constantly vigilant about non-endemic and rarely encountered diseases as well. The need for medical microbiologists to enable prompt diagnosis of such diseases cannot be overemphasized.

~ References


~ Figures
Cutaneous leishmaniasis (also known as oriental sore, tropical sore, chiclero ulcer, chiclero's ulcer or Aleppo boil, "Delhi Boil") is the most common form of leishmaniasis affecting humans. It is a skin infection caused by a single-celled parasite that is transmitted by the bite of a phlebotomine sandfly. There are about twenty species of Leishmania that may cause cutaneous leishmaniasis.