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Original Research Article

One-year prospective study of pediatric cutaneous leishmaniasis: a neglected tropical disease in sub Himalayan region, India

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ABSTRACT

Background: Satluj Valley of Himachal Pradesh is a newly identified focus of Cutaneous Leishmaniasis (CL) and here disease is still in emerging phase. Although children are most commonly affected victims by CL but till date no such study has been done on childhood CL in this region.
Methods: All newly diagnosed cases of CL were registered and detailed clinico-epidemiological parameters of children between 0-18 years were recorded. Imprint smears for LD bodies and biopsies for histopathology were done in atypical cases to confirm clinical diagnosis.
Results: Over a period of one year 337 cases of CL were registered, out of them 115 children (0-18 years) were enrolled for this study. School going (6-18 years) population was predominantly affected age group. Males outnumbered the females with a ratio of 1.88. Face was most commonly affected site and most (65.21%) cases presented with single lesion. Nodules (40.87%), plaques (24.35%) and papules (7.39%) were commonly observed morphologies although mixed patterns and atypical forms were also found. Most (62.60%) cases presented within 1-3 months of lesion onset. All cases responded well to treatment with antimonials. Lesional pain, swelling, bleeding, pigmentation and scarring were minor post treatment side effects.
Conclusions: Pediatric CL is an emerging infection along the Satluj valley of Himachal Pradesh (HP). Although clinical diagnosis is simple, and all cases respond well to antimonials. But, availability of diagnostic tests is essential to diagnose atypical cases. Further annual record keeping, and reporting is recommended to know the exact disease burden so as to promote the effective treatment and preventive measures.

Keywords: Cutaneous, Leishmaniasis, Pediatric, Sodium stibogluconate

INTRODUCTION

Cutaneous leishmaniasis (CL) is a major public health problem in some foci of Africa, Asia, Europe and North and South America.[1] Disease is endemic in approximately 98 countries across the world.[2] As per WHO, 1 to 1.5 million new cases of CL and 400,000 to 600,000 new cases of VL are detected every year.[3] The global prevalence of the disease is 12 million cases and 350 million people are considered at risk of contracting the infection.[4] In India, disease is prevalent in specific areas of Bihar, West Bengal, Uttar Pradesh, Assam, deserts of Rajasthan and the foothills of Sikkim.[5] CL affects various age groups depending on the infecting *Leishmania* species, geographic location, host specific immunity and disease reservoir.[1] In endemic areas children are at greater risk of acquiring disease than adults due to malnutrition and more exposure to vectors.[1]

In certain endemic areas it is seen that 60-70% of CL affected population is composed by 0-19 year children, thus emphasizing the importance of evaluating this population in more detail.[6]

There are about 20 species of *Leishmania* that may cause diseases in the form of cutaneous, muco-cutaneous and visceral leishmaniasis.[7] In old world, the CL primarily caused by *Leishmania tropica* in urban areas (dry type) and *Leishmania major* (wet type) in rural areas. Disease is transmitted by the bite of infected sand fly. After dermal inoculation, the leishmania parasite is up taken by dermal macrophages where further replication takes place. A raised, red shiny lesion develops at the site of the bite after an incubation period of 2-4 weeks. As per species the lesion either ulcerates and become secondarily infected or often spontaneously heals with atrophic scarring (*L. major*). Lesions of some leishmania species may re-appear *(*as satellite lesions) after healing of primary lesion (*L. viannia braziliensis)*.[7]

CL typically appears on exposed/uncovered body sites like face, neck, hands, feet and forearms. In children face being the most unprotected and exposed, is the commonest site for sand-fly bite.[8] Although CL is self-limiting and does not cause mortality, but it causes cosmetic disfiguration, persistence and spread into muco-cutaneous form thus increases expenses on treatment and side effects of available drugs.[9] Therefore, treatment is recommended to prevent these complications especially in children. The present study is aimed to encompass the whole clinico-epidemiological spectrum of the disease in children of Satluj Valley of Himachal Pradesh.

METHODS

All clinically diagnosed new cases of CL were registered from August 2016 to July 2017. Data of children between 0-18 years was extracted out from the registers on to the master chart. Demographic profile including age, sex, address, locality, family history and history of high risk activities (keeping domestic animals, farming/horticultural activities, outdoor playing) were recorded.

Clinical details about number, size, site, duration and type of lesion were also noted down. The diagnosis of CL was mainly based on the clinical criteria proposed by Kubba and AI- Gindan.[12] In typical cases treatment was initiated after clinical diagnosis while in atypical/doubtful cases, imprint smears for Leishman Donovan (LD) bodies and tissue smears for histopathology were taken.

As facilities for Novy-MacNeal-Nicolle (NNN) culture medium and PCR were not available in this institute so unproven cases were referred to higher centre for further workup. Thus, old cases, adults (>18 years) and unconfirmed cases were excluded out from this study.

All newly diagnosed cases of CL were started on antimonial treatment with intralesional (I/L) SSG on three alternate days in a week per month. Monthly follow up visits were conducted to note down the treatment response along with minor side effects.

RESULTS

Over a period of one year 337 patients of CL were registered, of which 115 children (0-18 years) were included in this study. In contrast to adults, amongst pediatric population males outnumbered the females by a ratio of 1.88. Affected age varied from 1.5 years to 18 years and mean age was 10.71 years. Most of the cases were from 7-12 years and 13-18 years age group (Table 1).

Table 1: Age and sex wise distribution of cases.

Age groups	Males		Females		Total	
	n	%	n	%	n	%
< 1 Year	0	0	0	0	0	0
1-6 Years	12	10.4	13	11.4	25	23.5
7-12 Years	30	26.1	17	14.7	47	40.0
13-18 Years	33	28.7	10	8.7	43	36.5
Total	75	65.2	40	34.8	115	100

All the cases were residing in prevalent foci lying close to Satluj River and all localities were surrounded by abundant vegetation. History of cattle rearing was positive in 60% cases but exposure to cowshed areas increased this rate to 95%. Affected cases were from district Shimla 54.8% (Rampur, Sunni blocks), Kullu 26.95% (Nirmand and Aani block), Kinnaur 13.05% (Tapri and Bhava Nagar block). 5.2% cases were migrants from Nepal and Bihar (Figure1).

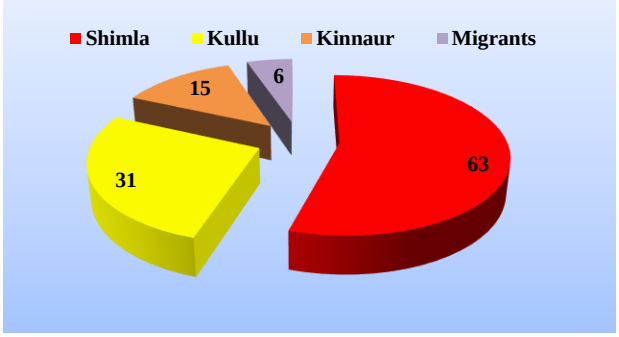

Figure 1: Area wise distribution of cases (n=115).

Majority (62.60%) of patients consulted doctor within three months of lesion onset, 18.26% presented between 4-6 months, 15.66% after 6 months and only 3.48% consulted doctor within one month of lesion onset. Patients were registered throughout the year, but case load was comparatively higher in the month of September

(16.5%), October (12.17%) and lower in the month of December (1.74%) and January (5.21%).

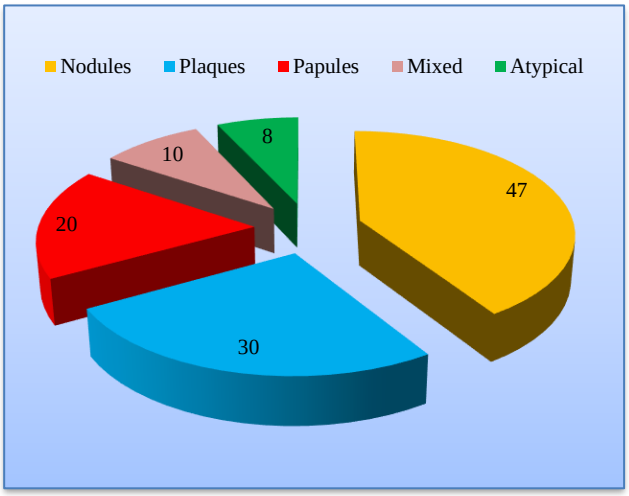

Figure 2: Morphological patterns of CL in children (n=115).

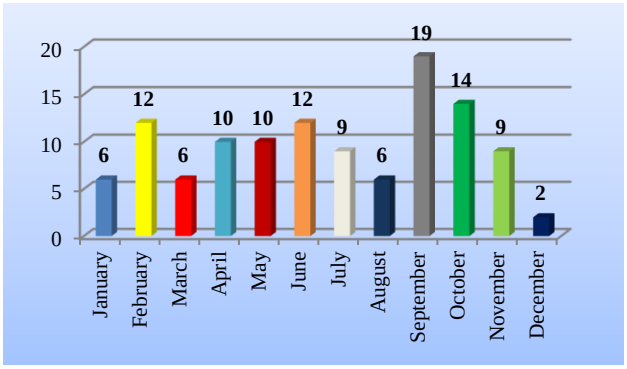

Figure 3: Month wise distribution of cases (n=115).

Family history was positive in 14(6.30%) cases. In total, 178 lesions were present in 115 patients, with the mean number of lesions being 1.54 per patient. Sizes of the lesions varied from 0.50 to 4 cm with mean size of 1.5 cm. Single lesions were most commonly seen in 65.21%, whereas 20% had two lesions, 12.17% had three lesions and multiple lesions were recorded in 2.26% cases. Common clinical variants were nodules (40.87%), plaques (26.11%) and papules (17.39%). Mixed variants like papulo-plaques, ulcerated plaques, plaques with satellite lesions and diffuse swellings were also observed. 8(6.95%) cases had atypical lesions including cheilitis, perleche, furunculoid, chancriform and verrucciform types.

Face was the most commonly affected site in almost every case. Lesions were predominantly present on cheeks (39.1%), chin (14.78%), nose (13.04%) and muco-cutaneous areas (lips and angles of mouth). Upper limb, lower limb and trunk were affected in 17, 1 and 1 cases respectively, whereas multiple sites were involved in 22.6% of cases (Table 2).

Imprint smears for parasitological examination and skin biopsies for histopathological assessment were performed in 8 cases showing atypical morphologies. Out of 8 patients LD (Leishman Donovan bodies) were detected on imprint smear in 5 (62.5%) patients. Histopathology reports revealed chronic grannulomatous inflammation comprising of macrophages, lymphocytes, epitheloid and plasma cells in all cases with intracellular LD bodies only in 4 (50%) cases.

Table 2: Distribution of lesions over body sites.

Site of lesion	n	%
Face (n=93)		
Forehead	6	5.2
Cheek	45	39.1
Eyelid	4	3.5
Nose	15	13.0
Chin	17	14.8
Jaw region	1	0.9
Submental region	5	4.35
Neck	6	5.21
Mucocutaneous areas (included in face) (n=28)		
Upper lip	19	16.52
Lower lip	5	4.35
Angle of mouth	4	3.48
Upper limb (n=17)		
Hands	1	0.87
Arm + Shoulder	11	9.56
Forearm	5	4.35
Lower Limb		
Legs	1	0.87
Trunk (n=1)		
Chest	1	0.87
Multiple (>1) body area involvement by multiple lesions	26	22.6

All cases were treated with 0.5-5 ml (100 mg/ ml) of SSG per lesion on three alternate days per month (one cycle). In most (59.13%) cases complete healing was observed by the end of 1st month, 33.9% showed resolution of lesion after 6 injections, 5.22% after 9 injections and 2 cases took 4 months for complete cure. Immediate side effects of intralesional SSG were local pain, swelling and minor bleeding in most of cases. Other side effects were secondary bacterial infection, ulceration, pigmentation, scarring, milia formation and reactionary submandibular lymphadenopathy.

DISCUSSION

CL can affect all age groups by protean cutaneous presentations. Although the incidence of CL is higher among pediatric population, but studies are sparse, and this is the first study on Pediatric CL from Himachal Pradesh. The reason for the greater incidence of CL in children is that children are exposed to the parasite at an early age, when there is lack of CL specific immunity.[10]

In present study out of total 337 registered cases, 115 children were affected by CL, thus they constituted 34.12% of the disease burden. Bari et al found childhood CL in 35.5% cases while Gurel et al from Turkey reported 19.7% cases in the 0-4 year age group of the total CL cases.[11,12] In endemic regions 60-70% of disease burden is composed by childhood cases. This shows that pediatric population is frequently affected by CL although prevalence can vary in different regions. Similar to other studies children between 7-18 years were predominantly affected age group in present study.[11] Higher predilection in this age group can be explained by frequent outdoor activities (school going, outdoor playing activities) thus making them prone to bites by infected vectors. Authors found a male to female ratio of 1.88 similar to studies by Agrawal, Bari and Shoaib et al.[8,11,13]

The lesions were most frequently seen on the face and neck (86.09%) in accordance with studies by Aksoy et al and Agrawal et al.[6,8] Face was the most commonly affected site in 80.87% cases in accordance with study by Zaraa et al.[14] Although a single lesion was most commonly observed, multiple lesions (more than one) were not uncommon. Bari et al observed multiple lesions in 25% of cases, which was similar to the finding in present study (27.8%).[11] Talari et al reported multiple lesions in 48.7% of cases, which is much higher than other studies and Zaraa et al reported multiple lesions in 8.4% of pediatric cases, which is lower than all other studies.[9,14] Family history of CL was present in 10.6% of cases in this study, whereas Kharfi et al and Qasmi et al reported it as 5.6% and 15.3% of cases, respectively.[15,16]

Diagnosis of CL is usually delayed due to painless and insidious nature of the disease. Average time lag between appearance of CL lesion and the first diagnosis have been reported as 12.71 months and 8 months by Zaraa and Kharfi respectively.[14,15] Authors found a shorter time interval of 3.19 months which indicates more disease awareness and early consultation by parents. The lesions were predominantly plaques and papulo- nodules and this has been reported in other studies also.[11,14,15] Uncommon morphological forms of CL like perleche, chancriform, erysipeloid and lupoid CL were observed in present study and they have been reported in literature also.[9,17]

In endemic areas, the clinical diagnosis is not difficult. Usually, this is best achieved clinical examination and further confirmed by performing imprint smear and histopathological examination. In present study imprint smears and histopathology were confirmatory in 62.5% and 50% of cases, respectively. Agrawal et al found imprint smear positivity in 70% cases and tissue smear positivity in 55.5% cases.[8] Aara et al found imprint smear positivity in 69.5% and tissue smears positivity for Leishmania parasites in 45.8%.[18] Whereas Rodrigues and Al-Hucheimi et al reported it as 66.7%, 66.2% and 66.7% and 59.6%, respectively.[19,20] Poor lab facilities and lack of regular pathologist in present lab was supposed to be the reason for lower positivity of present results in comparison to other studies.[8] Parasite culture in NNN media and species identification by PCR (newer DNA-based technique) have higher sensitivity but these investigations were expensive and not available in present institute.[21]

Currently intralesional sodium stibogluconate is considered as gold standard treatment for CL because of its efficacy and safety profile. Although it can be given by intramuscular and intravenous routes but significant adverse effects like pancreatitis, hepatitis, marrow suppression and QT prolongation have been reported.[7] The exact mechanism of action is unknown, but it is supposed that it inhibits the ATP synthesis. The World Health Organization recommends one intralesional injection of 1- 3 ml SSG repeated at 2-days intervals until complete subsidence of the lesion.[22] Aara et al followed a schedule of once or twice weekly intralesional injections till 5-7 doses.[18] While Sharma et al used intralesional SSG (1-5ml) on three alternate days per month.[23] Authors followed the later schedule due to logistic problems of the patients and further dose was adjusted according to the size of the lesions. All patients responded well to treatment except those who didn't complete the recommended cycles of injection SSG and later on presented with recurrence of lesion at original site. Complications were observed but they were short term and minor. Major problem was non-compliance by children due to intolerable pain during intralesional injection of SSG. This problem was overcome by anaesthetizing the lesion with I/L lignocaine prior to infiltration of SSG.

CONCLUSION

Childhood population makes a major portion of CL in Himachal Pradesh and its clinical spectrum is different from adult CL. Diagnosis in the pediatric age group is based predominantly on clinical presentation and history of stay in endemic areas. Invasive procedures are difficult to perform in pediatric cases and further there is low sensitivity of skin smear and biopsy. Intralesional SSG remains the standard treatment due to its efficacy, easy availability and minimal side effects.

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Ethical approval: The study was approved by the Institutional Ethics Committee

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