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Original Article

Socio-cultural and environmental risk factors for leprosy in Jaffna District, Sri Lanka.

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Abstract

This study was conducted to determine the socio-cultural and environmental risk factors of leprosy in Jaffna. A case-control study was conducted in dermatology clinic, teaching hospital, Jaffna from July 2015 to July 2016. All 97 cases and 291 controls by systematic sampling were recruited using interviewer-administered questionnaire.

The mean age of cases was 41 (SD=21) and control was 48 (SD=20). Of 97 cases, 54.6 %(n=53) males; 99 %(n=96) Tamils; 79.4 %(n=77) Hindus; 60 %(n=59) above secondary education. Of 291 controls, 42.3 %(n=123) were males; 99.3 %(n=289) Tamils; 85.6 % (n=249) Hindus; 78.4 % (n=225) above secondary education.

Socio demographic factors (12 variables), house and environmental factors (11 variables), and behavioral factors (18 variables) were included in bivariate analysis and many were found to be significantly ($p<0.05$) associated with leprosy. Marital status, displacement, monthly income, presence of BCG scar, firewood as cooking fuel, and hunting reptiles were not found to be associated with leprosy.

Multiple logistic regression analysis showed significant association between leprosy and - crowded household [OR4.5, (1.5-13.6)], non-availability of drinking water within 30 minutes walking [OR15.1, (2.3-98.3)], environment with shrub [OR2.9, (1.4-6.1)], exposure to shrub [OR2.7, (1.2-5.9)], exposure to leprosy patient [OR10.6, (4.1-27.5)], exposure to fishing [OR4.5, (1.5-13.6)], visits to leprosy affected area [OR10.8, (4.7-24.5)] and unavailability of safe water source [OR3.3, (1.2- 9.1)].

In addition to the well-known risk factors environment-related risk factors, especially exposure to jungle/shrubs could play a role in the transmission of leprosy.

Keywords

Leprosy, risk factors, Jaffna, Sri Lanka

Introduction

Leprosy is an ancient chronic communicable disease, caused by the bacterium *Mycobacterium leprae*. The global prevalence of leprosy decreased from more than 5 million cases in the mid-1980s to less than 200 000 by 2015 following the introduction of multidrug therapy (MDT) as a treatment for leprosy (1).

The available international, national, and sub-national data suggest that the distribution of leprosy cases is related to socio-economic and demographic factors that promote the disease spread and reduce the cure rate (2-5). Airborne diseases with long incubation periods like tuberculosis and leprosy have a strong relationship with environmental factors and standard of living. In 2006, Kerr-Pontes and his colleagues described the relationship between socio-cultural risk factors and leprosy (3).

In 1980, Sri Lanka adopted early diagnosis and complete treatment with MDT as the key strategies for reducing the disease burden due to leprosy (2). Sri Lanka was a pioneer in Asia to bring in multidrug therapy (MDT) in 1980 and full coverage was achieved in the same year. Sri Lanka achieved elimination status in 1995 (3). Since then Sri Lanka has been struggling to reduce the prevalence of leprosy much below the elimination target of one case per 10,000 population, with pockets of long-time leprosy endemic areas and some people seeking treatment late. The number of new cases has been increasing in recent years. Awareness regarding the disease remains low among the public resulting in delayed diagnosis (4).

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A similar situation has been observed in Jaffna District (population - 622,709) as well. More than half of the total cases in the district were reported from one Medical Officer of Health (MOH) area out of the 14 MOH areas in the district. Over the past 10 years, the number of cases reported or detected also showed a marked increase, and cases appeared in new geographic areas of the district (all MOH areas) with late presentations including all ages (more child cases) and both sexes (5). The present study was undertaken to determine the socio-cultural and environmental risk factors of leprosy in the Jaffna District, Sri Lanka.

Methods

Setting and participants

This case-control study was conducted from July 2015 to July 2016 with the cases and controls recruited from the dermatology clinic, at Teaching Hospital Jaffna. All leprosy cases (new and those who attended for follow-up during the study period) confirmed by a dermatologist were defined as cases. These included self-referrals as well as cases referred from house-to-house visits or mobile clinics. Other patients attending the dermatology clinic were selected as controls. Measures were taken to recruit as controls patients with diverse diagnoses.

Persons (both cases and controls) who stayed temporarily in the district and those who were unable to give the necessary information due to physical or mental infirmities were excluded.

Three controls were recruited for each case to increase the power of the recent data showed that about 39% of students in the district fail to qualify (GCE A/L result) for higher education (District Secretariat, 2014). We assumed that poor education level increased the risk of leprosy by about 1.9 times (3). The required sample size to detect this increased risk with 80% power and a level of significance of 0.05 was 103 cases and 309 controls.

All cases who attended the dermatology clinic (the cases included those who were being followed up at the clinic for leprosy treatment and those who came for the first follow-up visit after completion of treatment) during the study period were recruited. For each case, three controls were recruited on the following clinic day. The leprosy clinic is held on every Wednesday. On every Thursday, controls were recruited for the previous day's cases (if 3 cases were recruited on Wednesday, 9 controls were recruited on Thursday). The first control was selected randomly out of the first 10 patients registered on the particular day and consecutive patients were selected up to the required number. If the consecutive controls had the same diagnosis they were skipped and a patient with a different diagnosis was recruited to maintain the diversity of controls.

Instrument and data collection

A pre tested interviewer administered questionnaire (and interview guide) was used to collect the data. It consisted of socio cultural risk factors, demographic factors, environmental factors, behavioural factors and BCG vaccination status.

Pre-test was done in Kilinochi District. The patients' contact information was traced from the district leprosy register and data collection was done at home with the help of the district leprosy public health inspector (PHI). PI and one pre-intern doctor collected the data. Needed corrections were done in the questions mainly in wording and options (answers) of responses.

The data collectors (2 pre-intern doctors and one nursing graduate) were provided with adequate training prior to data collection to minimize interviewer bias and to ensure quality of data. Interview Guide was prepared by the PI and handed over to the data collectors before their training on data collection.

The training was done by the PI lasting a full day. A detailed explanation of the research objective and the related variables were explained to them. The PI discussed the data collection instrument in detail to ensure an understanding of the subject to be measured by the variables. Specific instructions for filling individual questions of the questionnaire were provided to them and the interview technique was demonstrated by role play among them. They filled out the first questionnaire (case) under the direct supervision of the PI on the same day. Following the training day, each data collector filled out four questionnaires (controls) in front of the PI and ensured the quality of data collection.

The selection criteria for the recruitment of controls described as in sampling section were explained. At the end of the session, the PI clarified the issues raised by the data collectors. Every week PI did random checks of 3-4 questionnaires to ensure completeness and gave appropriate feedback to data collectors.

Patients were interviewed after the completion of their clinical care. Interviews were conducted in a separate place without the interruptions of clinic staff after getting informed written consent. Some missed patients (who did not have adequate time to interview at the clinic) were interviewed at home by the same trained data collectors with the help of the area PHI or public health midwife (PHM) with prior arrangements.

Data analysis

During the collection, filled questionnaires were examined for completeness and missing data was attended at that point itself if the patient was available.

Coding of data was done by either the coordinator of data collection team or PI. Data was entered in to the computer and analyzed using SPSS version 21and WinPepi software. Entered data was re checked for any mistakes in data entry by cross checking and were corrected if found.

Univariate analysis was done for all the variables and background information were presented as simple descriptive statistics. Bivariate analysis was done to identify associations and described as crude odds ratio and 95% confidence interval with relevant p values.

The significant variables in the bivariate analysis were analyzed in forward stepwise logistic regression and presented as adjusted odds ratio and 95% confidence interval with relevant p values. Population Attributable Risk (PAR) with 95% confidence interval was calculated using the adjusted OR to each (individual) risk factor.

Ethical considerations

Permission was obtained from the Regional Director of Health Services, Jaffna and the director of the hospital. Ethical clearance was obtained from ethical review committee Faculty of Medicine, University of Jaffna.

During the data collection, voluntary participation was encouraged. From all the participants or guardians informed written consent was obtained. The interviewer read and explained both information sheet and consent form to illiterate participants in front of a witness (the accompanied person or Health Service Assistant or Nursing Officer. Parental Permission with Assent was obtained from 12-18 years old participants.

Results

Background

All 97 cases and 291 controls were recruited (94.2% of the required sample size). Mean age of cases was 41 (SD=21) years and controls was 48 (SD=20) years (table 1).

Table 1: Background of cases and controls

Description	Case (n=97)	Control (n=291)
Mean (and SD) age	41 (21)	48 (20)
Sex: male (%)	54.6	42.3
Ethnicity: Tamils (%)	99.0	99.3
Religion: Hindus (%)	79.4	85.6
Education: above secondary education (%)	60.0	78.4

Socio-demographic factors and leprosy

Among the socio demographic factors, age (OR=2.42), male sex (OR=1.65), low education level (OR=2.30), being unemployed (OR=2.10), child death in the family (OR=2.84), marriage of family members outside (OR=2.56), leprosy patient in marital place (OR=16.19) had statistically significant relationship with leprosy. Marital status, skin colour, displacement average monthly income did not have statistically significant relationship with leprosy (table 2).

Household and environmental factors with leprosy

All the studied household and environmental factors such as no electricity (OR=3.41), no water sealed toilet facilities (OR=2.61), use of dung or earth as flooring (OR=2.93), no safe water source within the house premise (OR=2.11), no access to safe water (OR=2.09) non-availability of drinking water within 30 minutes of walking distance (OR=7.44), environment with jungle/

able 2: Relationship of demographic factors with leprosy

Socio demographic factors		Case	Control	Odds ratio	95% Confidence Interval		p value
					Lower	Upper	
Age in years	0-15	14	19	2.42	1.16	5.03	0.018
	>15	83	272				
Sex	Male	53	123	1.65	1.04	2.61	0.035
	Female	44	168				
Education level	No/Primary	38	63	2.30	1.40	3.77	0.001
	Secondary/above	59	225				
Employment	Occupied	62	133	2.10	1.31	3.38	0.002
	Not occupied	35	158				
Marital status	Never married	36	19	1.42	0.88	2.31	0.152
	Ever married	61	272				
Skin colour	Dark	29	67	1.42	0.85	2.37	0.181
	Non dark	68	223				
Displacement	No/one time	41	112	1.16	0.73	1.85	0.541
	>one time	97	177				
Displacement within past 30 years	Yes	83	246	1.04	0.54	1.99	0.915
	No	14	43				
Child death in the family	Yes	13	84	2.84	1.30	6.20	0.009
	No	84	275				
Average monthly income	0-10,000	34	115	0.81	0.50	1.30	0.381
	> 10,000	63	172				
Marriage of family members outside	Yes	63	121	2.56	1.59	4.13	<0.001
	No	34	167				
Leprosy patient in marital place	Yes	8	2	16.19	3.20	81.93	<0.001
	No	21	85				

Table 3: Relationship of household and environmental factors with leprosy

Household and environmental factors		Cases	Controls	Odds ratio	95% Confidence Interval		p value
					Lower	Upper	
Electricity in household	Not available	17	17	3.41	1.67	6.99	0.001
	Available	80	273				
Water sealed toilet	Not available	15	19	2.61	1.27	5.36	0.009
	Available	82	271				
Dung or Sand in floor	Present	49	75	2.93	1.82	4.72	<0.001
	Absent	48	215				
Safe water source	Not available	83	214	2.11	1.13	3.93	0.019
	Available	14	76				
Access to safe water	Not available	65	143	2.09	1.30	3.38	0.003
	Available	32	147				
Drinking water access within 30 minutes' walk	Not available	7	3	7.44	1.89	29.37	0.004
	Available	90	287				
Environment with jungle/shrub	Present	61	52	7.76	4.66	12.91	<0.001
	Absent	36	238				
Satisfactory sewage disposal	Sometime	55	126	1.75	1.10	2.78	0.019
	Always	41	164				
Current house-hold crowding(persons/ room)	4/more	25	19	4.95	2.58	9.49	<0.001
	1-3 persons	72	271				
House-hold crowding 10 years back(persons/room)	4/more	32	47	2.55	1.50	4.31	<0.001
	1-3 persons	65	243				
Animals in house/yard	Sometime present	89	234	2.66	1.22	5.81	0.014
	Absent	8	56				

shrub (OR=7.76), poor sewage disposal (OR=1.75), crowded household (OR=4.95), house-hold crowding for 10 years (OR=2.55), availability in animals in house/ yard (OR=2.66) had statistically significant relationship with leprosy (table 3).

Behavioural risk factors

Among the behavioural factors, work in forest (OR=3.53), work in agriculture (OR=1.90), eating reptiles (OR=2.30), fishing (OR=8.41), exposure to open water source (OR=2.67), poor practice of hand washing (OR=3.06), exposure to jungle/ shrub (OR=4.44), live in jungle/shrub (OR=2.69), frequency of jungle exposure (OR=2.96), exposure to leprosy patient (OR=28.89), visit to leprosy affected area (OR=15.05), frequency of exposure to leprosy affected area (OR=6.21), experienced of food shortage (OR=3.35), frequency of changing bed linen (OR=1.75) had statistically significant relationship with leprosy. Other factors such as presence of BCG scar, firewood as cooking fuel, hunting in life time and hunting reptiles did not have statistically significant relationship with leprosy (table 4).

Multivariate analysis of significant risk factors associated with leprosy

All the statistically significant variables of the bivariate analysis, were analysed by forward logistic regression. Among them, crowded household (OR=4.54), no access to drinking water within 30 minutes' walk from home (OR=15.05), environment with jungle/shrub (OR=2.92), exposure to jungle/shrub (OR=2.72), exposure to leprosy patient (OR=10.63), exposure to fishing (OR=4.53), visits to leprosy affected area (OR=10.83) and unavailability of safe water source (OR=3.26) were found to be statistically significant in the final model (table 5). The highest PAR was found for visits to leprosy affected area (80.3%), followed by environment with jungle/shrub (54.8%), exposure

to leprosy patient (51.8%), unavailability of safe water source (44.9%) and exposure to jungle/shrub (36.7%).

Discussion

Among the statistically significant variables in the final logistic regression model, highest PAR was found for visits to leprosy affected area (80.3%), followed by environment with jungle/shrub (54.8%), exposure to leprosy patient (51.8%), unavailability of safe water source (44.9%) and exposure to jungle/shrub (36.7%). Environmental related risk factors found to be associated with leprosy except exposure to leprosy patient directly.

Some of the known risk/protective factors(6–9) environmental, and behavioural factors associated with risk of leprosy occurrence in the endemic North-eastern region. A case-control study in four municipalities on cases of leprosy diagnosed in the previous 2 years, with no other known, current, or past case of leprosy in the household or in the neighbourhood. Controls were individuals presenting for reasons other than skin problems to the health unit where the case was diagnosed and who lived in the same municipality as the case with whom it was matched. Low education level, ever having experienced food shortage, bathing weekly in open water bodies (creek, river and/or lake of leprosy did not show significant association in our study. This is not due to inadequate power of the study but due to high prevalence (>90%) of such factors like housing conditions, BCG coverage, etc. in Sri Lanka(10). Though Jaffna District was identified as a low risk area for leprosy some time ago and the fluctuating pattern of annual new case detection, the power of the study was increased by 2 methods: increased case: control ratio (1:3) and achieving an adequate sample size by recruiting prevalent cases who were diagnosed in previous year and being followed up during the study period.

Nearly similar picture was demonstrated in other studies also. A case-control study done in Brazil had shown that Low education level, ever having experienced food shortage, bathing weekly in open water bodies (creek, river and/ or lake) 10 years previously, and a low frequency of changing bed linen or hammock (more than biweekly) of current practices were significantly associated with leprosy(6)environmental, and behavioural factors associated with risk of leprosy occurrence in the endemic North-eastern region.

Table 4: Relationship of Behavioural factors with leprosy

Behavioural factors		Cases	Controls	Oddsratio	95% Confidence Interval		p value
					Lower	Upper	
Work in forest	Ever work	21	21	3.53	1.83	6.80	<0.001
	Not work	76	268				
Work in agriculture	Ever work	42	83	1.90	1.18	3.06	*0.008*
	Not work	55	207				
Hunting in life time	No Hunting	93	281	0.75	0.22	2.48	0.630
	Ever Hunting	4	9				
Hunting reptiles	Not hunt	93	281	0.99	0.26	3.75	0.992
	Ever hunt	3	9				
Eating reptiles	Ever eaten	23	35	2.30	1.28	4.13	*0.006*
	Never eaten	73	255				
Fishing	Ever fishing	29	14	8.41	4.21	16.78	<0.001
	Never fishing	68	276				
Exposure to open water source	Ever exposed	49	84	2.67	1.66	4.31	<0.001
	Never exposed	45	206				
Firewood as cooking fuel	Never used	3	16	0.55	0.16	1.92	*0.345*
	Ever used	94	274				
Hand washing	Poor practice	66	119	3.06	1.88	4.98	<0.001
	Good practice	31	171				
Exposure of jungle/shrub	Ever exposed	46	49	4.44	2.59	7.56	<0.001
	Never exposed	51	241				
Live in jungle/ shrub	Ever live	34	48	2.69	1.60	4.52	<0.001
	Never live	62	235				
Frequency of jungle exposure	More frequent	41	22	2.96	1.33	6.57	*0.008*
	Less frequent	17	27				
Exposure to leprosy patient	Some exposure	52	11	28.89	14.02	59.52	<0.001
	No exposure	45	275				
Visit to leprosy affected area	Possible visits	80	83	15.05	7.94	28.52	<0.001
	No visits	13	203				
Frequency of exposure to leprosy area	More frequent	58	35	6.21	1.91	20.22	*0.002*
	Less frequent	4	15				
Experience of food shortage at any time	Yes	69	123	3.35	2.04	5.50	<0.001
	No	28	167				
Frequency of changing bed linen	<Biweekly	68	166	1.75	1.07	2.87	0.026
	Biweekly	29	124				
BCG scar	Present	80	258	0.797	0.321	1.978	0.625
	Absent	7	18				

In addition, there are some other factors also contributing in different parts of the world. A study performed in Bangladesh found that younger age and close relationship with leprosy patients were independent risk factors for leprosy and presence of a BCG scar and sex were not risk for leprosy (11). A Malawi study also found no evidence of BCG status and associated risk for leprosy(12)000 initially disease-free individuals followed up during the 1980s in a rural district of northern Malawi. A total of 331 new cases of leprosy were diagnosed among them. Individuals recorded as living in household or dwelling contact with multibacillary patients at the start of follow-up were at approximately five- to eightfold increased risk of leprosy, respectively, compared with individuals not living in such households or dwellings. Individuals living in household or dwelling contact with paucibacillary cases were both at approximately twofold increased risk.

Table 5: Results of the logistic regression model of selected significant risk factors associated with leprosy

Exposure	OR (95%CI)	P value	PAR (95%CI)
Crowded Household	4.54 (1.51 to 13.61)	0.007	20.6 (10.3 to 29.6)
No access of drinking water within 30 minutes' walk from home	15.05 (2.30 to 98.34)	0.005	6.2 (0.8 to 11.4)
Environment with jungle/shrub	2.92 (1.40 to 6.06)	0.004	54.8 (41.1 to 65.3)
Exposure to jungle/ shrub	2.72 (1.24 to 5.98)	0.013	36.7 (23.0 to 48.0)
Exposure to leprosy patient	10.63 (4.10 to 27.59)	<0.001	51.8 (40.2 to 61.1)
Exposure to fishing	4.53 (1.51 to 13.60)	0.007	26.3 (15.9 to 35.5)
Visits to leprosy affected area	10.83 (4.77 to 24.59)	<0.001	80.3 (67.2 to 88.2)
Unavailability of safe water source	3.26 (1.17 to 9.05)	0.024	44.9 (7.2 to 67.3)

The higher risk associated with multibacillary contact and the fact that dwelling contact entailed a greater risk than household contact if the association was with multibacillary, but not with paucibacillary, disease suggest that paucibacillary cases may not themselves be sources of transmission, but rather just markers that a household has had contact with some (outside. But a study done in Brazil has shown that presence of BCG vaccination scar was a significant protective factor for leprosy (6)environmental, and behavioural factors associated with risk of leprosy occurrence in the endemic North-eastern region.

A study done in Bangladesh revealed that the food shortage and less dietary diversity with household food insecurity associated with an increased risk of having leprosy(15). Similarly, our study also had shown a significant association between food shortage and leprosy in bivariate analysis.

Several studies suggested person-to-person is not the only form of M. leprae transmission, and that indirect transmission might occur, and other reservoirs should exist outside the human body. A study done in Indonesia revealed M. leprae in water bodies as a possible source of infection in that area(16). Similarly a few studies discussed about soil as a possible source(17,18). Some studies explored variety of animals as source of M. leprae. Armadillos were one such animal species known as a source(19)Mycobacterium leprae. In the Americas,\r\nnine-banded armadillos (Dasypus novemcinctus. In 1985, it was proved that a certain type of monkey developed leprosy as naturally acquired infection(20). Likewise another study shown Red squirrels in the British Isles were infected with leprosy bacilli(21). Our study also revealed that a possible environmental reservoir playing a role in the transmission of leprosy in addition to person to person transmission.

As above, most of our findings are consistent with international data, multivariate analysis shown following are statistically significant risk factors in Jaffna district: crowded household, no access of drinking water within 30 minutes' walk from home, environment with jungle/shrub, exposure to jungle/ shrub, exposure to leprosy patient, exposure to fishing, visits to leprosy affected area and unavailability of safe water source. These factors being significant even after adjusting for the variables exposure to patient with leprosy and socio-demographic factors raises the possibility of an environmental source of infection for leprosy, may be an animal/ reptile reservoir being present either in land/water among jungle or shrub in the district. Future studies for the demonstration of this, will facilitate further reduction in case load and sustain a leprosy free nation.

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