Childhood leprosy: A retrospective descriptive study from Government Medical College, Kozhikode, Kerala, India

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Summary

Objective: To assess the profile and describe the clinical presentations and complications of childhood leprosy in a tertiary care hospital in North Kerala, South India during 2003–2012 and to analyse any change in the age-sex profile and the clinical pattern of leprosy in children below the age of 15 years over the 10-year study period.

Design: A retrospective descriptive study of children less than 15 years of age diagnosed with leprosy and registered for treatment in a tertiary care institution from 2003 to 2012. Demographic, clinical, investigative and treatment data were collected using a pre-set proforma.

Results: 138 (12·1%) of the total 1143 leprosy cases registered for treatment during the 10-year period were below 15 years of age. The 10-year study period witnessed a statistically insignificant decrease in the new childhood leprosy cases registered for treatment in our tertiary care institution. The majority of cases belonged to the 6–12 year age group (61·6%) with a male predominance. Borderline tuberculoid (BT) was the commonest clinical type (65·9%) followed by indeterminate leprosy (18·8%); 101 patients required paucibacillary (PB) and 37 needed multibacillary (MB) treatment. The number of patients requiring MB treatment showed a statistically significant increase and there was a significant decline in number of cases requiring PB treatment. During the entire study period no Type 2 lepra reaction was documented in patients below Hema 15 years and only two patients manifested Type 1 reaction. Ten (7·2%) out of the 138 patients were cases of relapse. There was a clear female predilection among relapse cases with the majority belonging to the adolescent age.

Conclusions: Childhood leprosy still contributes to a significant proportion of the total case load denoting the continuing active horizontal transmission of leprosy.

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The rise in number of patients with more extensive disease in the background of declining disease prevalence is suggestive of the delay in diagnosis and treatment. A high relapse rate noted in the present study may be due to incorrect classification and treatment of MB as PB leprosy which in turn might have resulted in treatment failure due to inadequate treatment.

Introduction

Elimination of leprosy as a public health problem is one of the major success stories of modern medicine. This was achieved at the global level in 2000 and India attained this status in January 2006.\textsuperscript{1,2} Leprosy elimination as a public health problem is defined as the reduction of disease prevalence to less than one per 10,000 population. In other infections elimination is defined as a reduction to zero of infection caused by a specified agent in a defined geographical area through deliberate efforts.\textsuperscript{2} If we use this criteria, leprosy is definitely not eliminated from the world.

With the declaration of ‘leprosy elimination’, leprosy services were integrated into the general health system. This was done with a view to ensure treatment to the affected at their nearest centre and to reduce the stigma associated with the disease. But recently Odisha, one of the first states in India to have disintegrated leprosy services reported a rise in prevalence of the disease.\textsuperscript{3}

Data based on the prevalence of registered cases for treatment may not reveal much about the current status of leprosy, as the shortened duration of fixed duration treatment (from 2 years to 1 year in multibacillary cases) in itself reduces the number of patients on treatment.

Moreover, it was often suggested that the rapid decline reported in number of leprosy cases from many parts of the world, was unlikely in a disease with such a long incubation period. It is possible that in order to achieve the elimination status, countries might have manipulated the statistics.\textsuperscript{4}

It is often suggested that childhood leprosy can serve as a better tool to assess the disease transmission in the society.\textsuperscript{5} Many studies in the post-elimination era documented a significant number of childhood cases including smear positive cases, pointing to the active disease transmission still taking place.\textsuperscript{5,6}

Hence we decided to conduct a retrospective descriptive study on leprosy among patients below 15 years who attended the outpatient department (OPD) the of Government Medical College, Kozhikode with cardinal features of leprosy from 2003 January to 2012 December.

Materials and methods

**STUDY DESIGN: RETROSPECTIVE DESCRIPTIVE**

This study was a retrospective analysis of all leprosy cases less than 15 years of age, who registered for treatment at the Dermatology department of the Government Medical College, Kozhikode from January 2003 to December 2012. This institution is a tertiary-care teaching hospital situated in the north of Kerala, a state in South India. Ethical clearance was obtained from the institutional ethics committee of Government Medical College, Kozhikode on 23.5.2013.
A diagnosis of leprosy was made, when a patient presented with any of the cardinal features of leprosy (asymptomatic hypopigmented or erythematous skin lesion with definite loss or impairment of sensation or thickened peripheral nerve with sensory impairment in the area supplied by the nerve or skin smear positive for acid fast bacilli).

A pre-set proforma was used to collect data regarding age, sex, possible source of contact, clinical findings and investigations from previous case records. A ‘household’ or ‘intra-familial’ contact was defined as any person with current or past history of leprosy in the immediate family (parents, siblings, grandparents) living in the same house and partaking in meals from a common kitchen. Known cases from the immediate neighbourhood of the patient’s house were considered as ‘extra-familial’ contacts. Clinical features including size, site, morphology and number of skin lesions were noted. Nerve function impairment (NFI), when present was charted out. Sensory impairment was detected by the inability or reduced ability to appreciate temperature, pain and fine touch. Test tubes containing water at 40°C and at 25°C were used to test temperature sensation, pain sensation was tested using pin prick and a wisp of cotton was used to check fine touch. Motor impairment was diagnosed when less than grade 5 power was recorded on voluntary muscle testing. Criteria by the World Health Organisation was used for disability grading.

From each leprosy patient one smear from an ear lobe smear and at least two more slit skin smears (from representative skin lesion and normal skin) were taken routinely in our institution and were stained with Ziehl-Neelsen technique to determine the morphological and bacteriological indices. Biopsies from the leprosy lesions were stained using haematoxylin and eosin to study the morphology and Wade Fite (modified Ziehl-Neelsen staining technique) to identify the acid fast bacilli (AFB).

Patients were categorised as per the Ridley-Jopling classification.

Patients who had only nerve lesions without any cutaneous manifestations and who satisfied the cardinal criteria for leprosy were placed under the category of neuritic leprosy and patients who manifested vague hypopigmented patches with doubtful sensory impairment and perivascular and peri-appendageal lymphocytic infiltration on histology were diagnosed as indeterminate leprosy.

Patients who had features of Type 1(T1R) or Type 2 (T2R) lepra reaction were classified accordingly. (A clinical diagnosis of T1R was made when a patient in the borderline spectrum of leprosy had acute onset of eryt and oedema of skin lesions with or without neuritis and oedema of the hands, feet and face. T2R was diagnosed when a BL or LL patient had crops of tender subcutaneous skin lesions with or without accompanying neuritis, iritis, arthritis, orchitis, dactylitis, lymphadenopathy, oedema and fever.

The disease spectrum as well as the treatment received were documented. Patients presenting with six or more skin lesions or two or more enlarged nerve trunks or skin smear positivity for acid fast bacilli were treated with multibacillary regimen and patients presenting with less than six skin lesions, less than two enlarged nerve trunks and a negative skin smear for acid fast bacilli were treated with paucibacillary regimen.

The number of patients who had developed Grade 2 disability at the time of initial presentation were documented. Grade 2 disability was defined as the presence of visible deformity or damage (ulceration, shortening, disorganization, stiffness and loss of part of or all of the hand or foot) affecting hands and feet due to leprosy or visual acuity less than 6/60 or inability to count fingers at a distance of six meters caused by leprosy.

Patients who attended the OPD with suspected leprosy relapses were noted and data regarding the previous disease spectrum and previous treatment received were documented.
Relapse in leprosy was diagnosed when a patient who successfully completed an adequate course of multidrug therapy, subsequently developed new signs and symptoms of the disease, either during the surveillance period (2 years for PB and 5 years for MB leprosy) or thereafter.\textsuperscript{5}

The data was studied with respect to the age and sex distribution, clinical features, complications and the treatment received by leprosy patients below the age of 15 years. The proportion of new childhood leprosy cases with respect to the total number of new leprosy patients who attended our institution during the 10-year period was also studied. The epidemiology of the new childhood leprosy patients who attended our institution over the decade (2003–2012) was analysed using chi-square test for linear trend using the StatCal component of the Epi Info version 7.1.2 and an attempt was made to detect any change in the pattern over the years.

Results

138 (12.1\%) of the total 1143 leprosy patients registered for treatment in our institution from 2003 to 2012 were below the age of 15 (Table 1).

The number of new childhood leprosy cases showed a statistically insignificant decline over the 10-year study period ($P$ value 0.16).

The majority of cases belonged to the 6–12 years age group (61.6\%, Table 2) which remained unchanged throughout the 10-year interval ($P$ value 0.301).

The age of the affected children ranged from 2 to 15 years.

There was a clear male predilection; (Table 2) no significant change was observed in the sex distribution of the affected over the years ($P$ value 0.163).

The commonest disease spectrum throughout the duration of the study was BT. Only 3-6\% of our patients belonged to mid borderline (BB), borderline lepromatous (BL) and lepromatous leprosy (LL) spectra (Table 3).

Table 1. Ten year distribution of new childhood and total leprosy cases in a tertiary care institution (2003–2012)

| Year | Childhood leprosy | Total number of leprosy cases | Percentage of childhood leprosy among total leprosy |
|------|-------------------|-------------------------------|-----------------------------------------------------|
| 2003 | 26                | 169                           | 15.4                                                |
| 2004 | 21                | 186                           | 11.3                                                |
| 2005 | 17                | 118                           | 14.4                                                |
| 2006 | 16                | 108                           | 14.8                                                |
| 2007 | 4                 | 84                            | 4.8                                                 |
| 2008 | 8                 | 110                           | 7.3                                                 |
| 2009 | 21                | 113                           | 18.6                                                |
| 2010 | 9                 | 75                            | 12.0                                                |
| 2011 | 9                 | 92                            | 9.8                                                 |
| 2012 | 7                 | 88                            | 8.0                                                 |
| Total| 138               | 1143                          | 12.1                                                |

$P = 0.16$ for comparing new childhood leprosy cases Versus new adult leprosy cases attending a tertiary care institution from 2003–2012
Seventeen patients (12.3%) gave a history of contact with a leprosy case; 15 of them were household contacts and two were family members living in a nearby house. The 2-year old who presented with BT had a household contact in his paternal grandfather who was receiving treatment for BL.

Of the 138, 73.2% required PB treatment and the rest received MB treatment [Figure 1]. The number of patients requiring MB treatment showed a statistically significant increase and there was a significant decline in number of cases requiring PB treatment (P value 0.009, Figure 2). 3.6% of the total were smear positive for AFB.

Enlarged nerves were detected in 45.7% (Figure 2) with 23.9% manifesting multiple nerve thickening. A statistically significant increase was noted in number of patients presenting with nerve involvement (P value 0.001, Figure 3) and an increase (though statistically insignificant, P value 0.16) was noted for those presenting with multiple nerve involvement.

Grade 2 disability was observed in eight patients. All were in the 13 to 15 age group. Seven were males. Four children suffered from clawing of fingers and foot drop developed in two. They were treated with steroids and physiotherapy and two patients had residual weakness. Two children at the time of diagnosis had trophic ulcers affecting the heel and the ball of the great toe respectively.

One of the male patients with Grade 2 disability developed nerve palsy as part of the neuritis of Type 1 reaction. Only one more patient (a 9-year old girl) developed Type 1 reaction during the study period that manifested as neuritis and ulcerated skin lesions (Figure 4).

During the entire study period no Type 2 reaction was documented in patients below the age of 15.

Ten patients (7.2%) were diagnosed as relapse cases. Seven of them had received multidrug therapy from other institutions and were referred to us when symptoms reappeared.
Table 3. Spectrum-wise distribution of childhood leprosy

| Year | IL M | IL F | IL Total | NL M | NL F | NL Total | TT M | TT F | TT Total | BT M | BT F | BT Total | BB M | BB F | BB Total | BL M | BL F | BL Total | LL M | LL F | LL Total | Childhood cases/year |
|------|------|------|----------|------|------|----------|------|------|----------|------|------|----------|------|------|----------|------|------|----------|------|------|----------|---------------------|
| 2003 | 7    | 3    | 10       | 0    | 0    | 0        | 1    | 1    | 2        | 10   | 4    | 14       | 0    | 0    | 0        | 0    | 0    | 0        | 18   | 8    | 26       |
| 2004 | 4    | 1    | 5        | 0    | 0    | 1        | 1    | 2    | 3        | 7    | 4    | 11       | 0    | 0    | 1        | 10   | 1    | 11       | 14   | 7    | 21       |
| 2005 | 1    | 0    | 1        | 0    | 0    | 0        | 0    | 0    | 0        | 11   | 4    | 15       | 0    | 0    | 0        | 1    | 1    | 2        | 12   | 5    | 17       |
| 2006 | 1    | 0    | 1        | 0    | 0    | 0        | 2    | 1    | 3        | 7    | 4    | 11       | 1    | 0    | 1        | 0    | 0    | 0        | 11   | 5    | 16       |
| 2007 | 0    | 0    | 0        | 0    | 0    | 0        | 1    | 1    | 2        | 1    | 1    | 2        | 0    | 0    | 0        | 0    | 0    | 0        | 1    | 3    | 4        |
| 2008 | 0    | 1    | 1        | 0    | 0    | 0        | 0    | 0    | 0        | 5    | 2    | 7        | 0    | 0    | 0        | 0    | 0    | 0        | 0    | 0    | 0        | 5    | 3    | 8        |
| 2009 | 1    | 3    | 4        | 0    | 0    | 0        | 1    | 1    | 2        | 7    | 8    | 15       | 0    | 0    | 0        | 0    | 0    | 0        | 0    | 0    | 0        | 9    | 12   | 21       |
| 2010 | 2    | 1    | 3        | 0    | 0    | 0        | 0    | 1    | 1        | 4    | 0    | 4        | 0    | 0    | 0        | 1    | 0    | 1        | 0    | 0    | 0        | 7    | 2    | 9        |
| 2011 | 1    | 0    | 1        | 0    | 0    | 0        | 0    | 1    | 1        | 5    | 2    | 7        | 0    | 0    | 0        | 0    | 0    | 0        | 0    | 0    | 0        | 6    | 3    | 9        |
| 2012 | 0    | 0    | 0        | 0    | 0    | 0        | 0    | 1    | 1        | 2    | 3    | 5        | 0    | 0    | 0        | 0    | 0    | 0        | 1    | 0    | 1        | 3    | 4    | 7        |
| 10 year Total | 17 | 9 | 26 | 1 | 1 | 2 | 5 | 9 | 14 | 59 | 32 | 91 | 1 | 0 | 1 | 2 | 1 | 3 | 1 | 0 | 1 | 86 | 52 | 138 |

IL = Indeterminate leprosy; NL = Neuritic leprosy; TT = Tuberculoid leprosy; BT = Borderline tuberculoid leprosy; BB = Mid borderline leprosy; BL = Borderline lepromatous leprosy; LL = Lepromatous leprosy; M = Male; F = Female
after the completion of treatment. All except one had documents of receiving regular treatment. One child did not have any details of previous treatment, but according to her parents received regular treatment for the recommended duration. Eight out of the 10 were girls, indicating a clear female predilection ($P$ value 0.004). One girl and one boy were 9 years and 10 years old respectively at the time of the relapse and the rest were 12 to 15 years of age. Eight out of the 10 were treated with PB MDT in the past and the interval between the

Figure 1. New childhood leprosy cases registered for PB and MB treatment in a tertiary care institution from 2003 to 2012.

Figure 2. Thickened nerve in childhood leprosy.
completion of treatment and the relapse varied from 3 months to 2 years. All relapsed cases manifested clinical features warranting re-treatment with MB MDT.

Discussion

Seven years after the declaration of the elimination of leprosy, childhood leprosy still contributes to a significant proportion of the total case load and this denotes that active horizontal transmission is still taking place. The childhood cases contributing to 12.1% of the total, noted in our study was much higher than the 4.5% reported by Burman et al., but was comparable to the findings in certain other studies.

The commonly affected age group remained 6–12 years which was consistent with the incubation period of the disease. Another study in the post-elimination era documented the majority of patients in the above 11 age group.

The fact that we could identify the contact case in only 17 of our patients points to the hidden cases in the community. Similar findings were reported in some studies but higher figures were noted in other studies. This suggests that even in low endemic areas, mass leprosy detection camps have a role to play.

Our observation of predominance of BT was in concordance with earlier studies. Compared to other studies our study documented less patients with extensive disease with respect to nerve involvement, treatment required (MB MDT), smear positivity and Grade 2 disability at initial presentation. But the number of patients requiring MB treatment and those with multiple nerve involvement showed a rise during the later years of the study.

The increase observed in cases requiring multibacillary treatment including those with multiple nerve involvement could be due to a waning of disease-specific immunity in the community that has taken place with the decline in disease prevalence (leading to extensive disease manifestation in the affected). The single LL case observed in the study group

Figure 3. Nerve trunk involvement in new childhood leprosy cases attending a tertiary care institution from 2003 to 2012.
Presented during the last year of the study. Another possible explanation is the delay in diagnosis that has been already reported from different parts of the world due to the diminishing expertise in detecting early leprosy lesions.4 With the declaration of ‘leprosy elimination’, there is a waning of awareness of leprosy among the health care workers and the general population.4 Experts had predicted that as the prevalence of leprosy comes down, the major challenge will be in making the correct diagnosis early, in those affected.4

5.8% of the study group developed Grade 2 disability at initial presentation. An increase documented in Grade 2 disability among the newly registered adult leprosy patients who attended our institution during the same interval was not observed in childhood leprosy.

The development of severe T1R in a 9-year old girl observed in our study highlights the need to look for this complication in childhood leprosy as a delay in diagnosis and treatment of the same can lead to irreversible nerve damage.

The majority of relapse cases in our study belonged to the 12 to 15 years age group as expected in a disease with a prolonged incubation period. There was a striking female predominance noted in relapse cases which was contrary to the sex profile observed in the total number of leprosy cases. The higher susceptibility of adolescent females to leprosy relapse may be attributed to the hormonal changes and the subsequent alterations taking place in the immune system with menarche.
Our study documented a higher relapse rate compared to some other studies;\(^5\) 80% of the relapses occurred following PB treatment and 70% had received previous treatment from other institutions. All 10 patients, on evaluation were found to have disease requiring retreatment with MB regimen. Probably a wrong classification of MB as PB disease and subsequent inadequate treatment might have contributed to the high relapse rate. One patient did not have any documents regarding previous treatment received, hence we could not rule out the possibility of irregular treatment.

All the relapsed cases in our study developed reappearance of symptoms within 2 years of completion of treatment. The major diagnostic challenge was in differentiating relapse from late T1R. None of them showed any clinical evidence of lepra reactions.\(^9\) One patient came to us with suspected relapse within 3 months of completion of treatment. This patient after completion of MDT complained of the appearance of a new skin lesion with sensory impairment and without any clinical evidence of lepra reaction. The appearance of a non-tender, non-erythematous new single skin lesion satisfying the cardinal criteria for leprosy in a treated patient was more in favour of a relapse than a lepra reaction as the latter is usually associated with multiple erythematous oedematous tender skin lesions. In spite of this, in view of the short interval between the completion of the treatment and the appearance of the new skin lesion, the patient was given a therapeutic trial with systemic steroids. Since the patient did not show any improvement after 4 weeks, and as the histopathology analysis of the new skin lesion revealed borderline tuberculoid leprosy with no evidence of dermal or intra-granuloma oedema, a final diagnosis of relapse was made rather than late T1R.\(^15\) The patient gradually responded to standard MB-MDT as she had enlargement of two nerve trunks (without clinical evidence of neuritis) in addition to the skin lesion.

The major limitations of our study was the small sample size (138 cases over 10 years) and the dependence on data collected from previous case records in a tertiary care institution. This data does not reflect the status of the disease in the general population, as the more severely affected patients usually seek advice in a tertiary referral unit.

**Conclusions**

Leprosy transmission still takes place as reflected by the significant portion of childhood cases. The rise in number of patients with more extensive disease in the background of declining disease prevalence highlights the need to reassess the efficacy of the existing system in early case detection. Diagnosing leprosy requires training. For the successful implementation of leprosy services provided through the general health system, training of health workers, regular refresher classes and continuous monitoring are essential. Health care workers should be proficient in identifying anaesthetic skin lesions and enlarged nerves, as about 30% of the MB cases will be missed if a patch with sensory impairment is taken as the single criterion to diagnosis leprosy.\(^15\) Inexperienced health workers may miss the early lesions of leprosy, leading to delay in diagnosis or may incorrectly classify and treat MB as PB leprosy, thus causing treatment failure. This incorrect classification could be the reason for the high relapse rate noted in the present study especially following PB regimen. But the female preponderance noted among the relapse cases cannot be ignored. We recommend that after completion of treatment, affected female children should be kept under follow-up throughout the adolescent years, as female sex and pubertal age group are found to be the major risk factors for relapse in our study.
It is of paramount importance to maintain the awareness among health care professionals and public that ‘elimination of leprosy as a public health problem’ does not necessarily imply the elimination of the infection. Most of the affected belonged to the ‘above 6 years’ age group, denoting the importance of school surveys in early case detection. Early case detection and treatment administration has played a pivotal role in reducing the disease burden in the past and ensuring the same is the need of the hour to prevent the re-emergence of this ancient disease.

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