

EDITORIAL

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The 6th WHO TAG Report: Validation and “Non-existent Patients”

The Report of the 6th World Health Organization Technical Advisory Group on Elimination of Leprosy contains much interesting material, and we strongly recommend that all ILA members and other interested persons read it carefully (WHO/ CDS/ CPE/ CEE/ 2004.41) (<http://www.who.int/lep/>). Section 11, entitled “Validation of diagnosis of newly detected cases,” particularly caught our attention.

This section reports the results of a study of the accuracy of the diagnosis of leprosy in clinics in several regions of India. The study was conducted by having two individuals with substantial clinical experience in leprosy review the patients initially evaluated by health care workers at the clinics, to ascertain the accuracy of the original diagnosis. The study reports, among other things, that over 30% of the patients in the Delhi clinics were “non-existent patients” because they could not be confirmed to live at the addresses given, which in some cases were fictitious addresses. Based on this and related information, the study concluded that leprosy was greatly over-diagnosed in these clinics, and a policy recommendation is made that such patients should be removed from the leprosy register.

This report and its recommendations are perplexing. First, what was done was not a true validation study at all, but the recording of a second (and third) opinion. This may sometimes be useful, but it is *not* validation. A true validation study requires assessment by an independent test or method, not merely asking an independent observer. In leprosy, three such independent means of assessment are readily available: skin smears, histopathological examination of biopsies, and application of polymerase chain reaction (PCR) to detect *M. leprae*

in appropriate specimens. Each of these has its own constraints, but when applied within a proper study design, these independent means of assessment can provide validation of the diagnosis within definable statistical boundaries. (It is notable that the most basic of these methods, biopsies and skin smears, are no longer available in many clinics around the world because the WHO, now concerned about the accuracy of the diagnosis, for several years has strongly recommended eliminating these tests from the programs.)

Reporting the main findings of the study, the authors note that: “*The proportion of non-existent cases was high in Delhi (31.3%). . . Non-existent cases are defined as patients who were on the list prepared by the local health facility, but who could not be found at the given address and whose existence was unknown to community/ neighbors in the area.*” For these patients, then, there was no opinion or evidence that the diagnosis was found to be incorrect. Rather, these are patients for whom the initial health worker made a diagnosis of leprosy, but the people could not be found later at the address given.

Two possible reasons for this finding are offered: “[1] *these were either fictitious cases or* [2] *individuals who gave a misleading address.*” This first explanation is favored by the authors of the report, who have then labelled these individuals as “non-existent patients.”

If this explanation is correct, i.e., that these are fictitious cases, it implies that nearly 1/3 of all patients in the Delhi clinics have spent most of a day uselessly queuing up in lines in hot, crowded clinics in order to obtain the diagnosis of leprosy, a diagnosis which brings them no gain but, rather ,

may consign them to lifelong stigma, ostracism, family rejection, and reduced employment and earning ability. Most of these people depend on each day's earnings to provide food for themselves and their families for that day. While it is possible that one or two deranged individuals may waste their days in clinics pretending to have leprosy, it is clearly illogical to conclude that nearly 1/3 of the patients in the Delhi clinics do so. Yet this report implies that this is the most likely explanation and then gives them the pejorative label of "non-existent patients." Finally, based on this conclusion, one of the main recommendations in the next section (1.1.2), is that: "*Non-existent patients should be removed from the leprosy register.*"

In contrast, the second explanation (i.e., individuals gave misleading addresses) is a phenomenon well-known to all leprosy control programs: individuals move to another city, another region, or even another country when they suspect the diagnosis, to avoid the possibility that friends or family will learn that they have leprosy. It is ludicrous to suggest that 1/3 of patients are "faking" leprosy, while it is obvious that these individuals are concealing the diagnosis from their own communities. These, then, are *not* "non-existent patients" but non-existent addresses for *real* patients needing medical care. This also indicates that there is much more work to be done, not less, in finding and treating those patients who gave fictitious addresses. If 1/3 of the patients are evading follow-up, it is highly likely that a substantial number of them are capable of transmitting the disease to their close contacts. Rather than expend energy on the avoidance of over-treatment, more energy must be expended in delivering appropriate treatment.

What can be done to avoid such over-interpretation of information and formulation of premature recommendations? One suggestion is that more caution be exercised in accepting evidence put forward, and in drawing conclusions from that evidence. This is one area in which medical and scientific journals can play a valuable role.

Everyone who has submitted a research report to the JOURNAL, or to any other peer-reviewed journal, is familiar with the rigorous process of review, requests to revise

and re-submit a paper to make it clearer, to improve its statistical treatment of the data, and to avoid over-interpretation of results and over-reaching for conclusions. In spite of its flaws, this process is regarded by the worldwide medical and scientific community as a necessary step in the larger process of ensuring that new treatments or treatment policies are based on sound evidence and that their implementation is ethical. Clearly, recommendations for treatment of leprosy should be given no less care and consideration.

An example of such an approach is the comprehensive set of recommendations based on careful examination of the evidence concerning diagnosis, chemotherapy, and epidemiology of leprosy, contained in the Report of the ILA Technical Forum⁽²⁾. The evidence presented in that report was discussed in many of the symposia and workshops of the 16th International Leprosy Congress, and after a spirited debate over several aspects of the recommendations, the ILA General Assembly passed a Resolution calling on "all stakeholders (including national governments, international organizations and non-governmental organizations) to *review their recommendations in the light of this report*" (emphasis mine)⁽¹⁾. This careful, measured approach still seems the wiser path.

The accurate diagnosis and classification of leprosy has always been essential in the delivery of the best care to individual patients and in formulating the best health policy in individual countries. In a previous issue we addressed concerns over the inadequacies of some classification schemes for research studies⁽³⁾. We now agree with the goal of the validation study described in the 6th WHO TAG report, and hope that a more careful study can be performed to address the important issues raised.

In the meantime, we hope that Ministries of Health around the world, and their leprosy control officers, will not be too eager to accept the Report's premature recommendations related to those patients who are not easily located. It would be most unfortunate to adopt policies to disregard these patients, both because the patients and their contacts are not well served by such a policy, and because the resulting numbers in the registry may then seriously under-

estimate the true extent of the disease. Certainly we should not condone the addition of any more pejorative labels such as “non-existent patients” to describe the worried, wary individuals whose trust the leprosy control workers must gain in order to give them the best medical care.

—DMS

REFERENCES

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