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EDITORIALS

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DIMORPHOUS MACULAR LEPROSY

A SPECIFIC CLINICAL APPLICATION OF "DIMORPHOUS"

An article entitled "Dimorphous Macular Leprosy," by Dr. S. G. Browne of the Yalisombo Leprosarium in the Belgian Congo, appears in this issue although no such variety of leprosy is recognized in formal classification. It was accepted for publication for two reasons. First, it deals with a peculiar form of macular leprosy occurring in Africa which apparently should be given recognition and a distinctive name (which should, preferably, also be appropriate) and should be known about and sought for elsewhere. Second, the application of the designation "dimorphous" to such a clinical entity may lead to clarification of a confused situation which has resulted from the way that term has been used in the past, first as an alternative for "borderline" and subsequently in other ways.

I. Ryrie, during a tour in southern Nigeria in 1947, was struck by a macular syndrome different from anything he had known in Malaya.[1] The condition usually started with a single primary macule, multiple macules being (probably) secondary as a rule. A "macular reaction" might occur in which an eruption of such lesions would appear almost overnight. He believed the lesions to be bacteriologically negative and without anesthesia; they were not to be confused with the "juvenile leprosy" of Muir or the "prelepromatous macule" of Cochrane.

[1] Ryrie, G. A. The macular syndrome in Nigeria. Leprosy Rev. **19** (1948) 35-39.

Two other reports also told of cases with peculiar "uncharacteristic" or "indefinite" macular lesions frequent in that part of Africa. Lengauer [2] said that in the Western Region of Nigeria most of the patients with lepromatous leprosy gave a history of a single depigmented macule, usually followed later by an outbreak of a fresh crop of ill-defined macules; lepromatous infiltration occurred much later. Among 185 bacteriologically negative cases examined, 37 per cent were "macular," with lesions described as "vague." Ross,[3] visiting Gambia, saw marked differences in leprosy there from Eastern Nigeria, including generally greater bacteriological positivity. Of the cases found, 15.5 per cent were of an "unclassified" kind, with "uncharacteristic" lesions whose color was often puzzling; 85 per cent were bacteriologically positive.

Thus it would seem that there is seen in West Africa a peculiar macular variety for which formal classification does not provide. Lowe was doubtful about that, however, because during one year's work in Nigeria he had seen only one case with macular lesions showing no anesthesia and no bacilli.[4] So far as we are aware the matter has not been pursued in that region, but it is now evident that such a form occurs not infrequently in the Belgian Congo.

II. Consideration of the present confusion of terminology concerning "borderline" and "dimorphous" calls for a bit of delving into history, first for recognition of cases with both tuberculoid and lepromatous characteristics, and second for certain recent vicissitudes of classification and terminology.

The first report of reactional changes in tuberculoid leprosy [5] told of several patients (South Africans) who, from the clinical appearance, had been thought to have undergone partial transformation to lepromatous ("nodular," Davison), especially with respect to the lesions of the face. Although the laboratory examinations that were made afforded no proof of that change, it was not doubted that "transformation from the tuberculoid to the lepromatous type (or to a mixed type) may occur."

Lowe, in Calcutta, soon recognized cases which appeared to be intermediate between tuberculoid and lepromatous, but he did not study them particularly or attempt to give them a distinctive name.

Contrary to common understanding, his "N?C" designation (which now would be T?L) was first applied to reactional tuberculoid cases,[6] which as is to be seen from his outstanding study of tuberculoid lesions [7] resembled those called *akuter Schub* by the

[2] LENGAUER, L. Leprosy in the Benin and Warri areas of Nigeria. Leprosy Rev. **19** (1948) 14-20.

[3] ROSS, C. M. Some differences in the leprosy of the Gambia and Nigeria. Leprosy Rev. **19** (1948) 12-14.

[4] LOWE, J. AND SMITH, M. The chemotherapy of leprosy in Nigeria. Internat. J. Leprosy **17** (1949) 181-195.

[5] WADE, H. W. Tuberculoid changes in leprosy. II. Lepra reactions in tuberculoid leprosy. Internat. J. Leprosy **2** (1934) 279-292.

[6] WADE, H. W. Regional variations of leprosy with special reference to tuberculoid leprosy in India. Leprosy in India **9** (1937) 3-13.

[7] LOWE, J. A study of macules in nerve leprosy with particular reference to the "tuberculoid" macule. Leprosy in India **8** (1936) 97-112; *reprinted in* Internat. J. Leprosy **5** (1937) 181-198.

Japanese. Cases actually of the borderline class were distinguished, in a joint report,[8] as "secondary cutaneous" ". . . with macules now or originally of nature of [tuberculoid] cases, and with slight or moderate [lepromatous] type changes, usually with few bacilli."[9] Only later did he say[10] that he "provisionally" classified such cases as N?C; and he did not apply that designation to the "intermediate type of case common in Burma which was so difficult to classify."[11]

Cases which had been tuberculoid but which seemed to have changed, as a result of reactions, *toward* the lepromatous state were specifically described and designated "borderline tuberculoid" by Wade and Rodriguez,[12] and in a further article Wade[13] called attention to an excellent example of borderline in a case which had been reported with pictures in 1893 by Arning and Nonne as "tubero-macular." The borderline form was later given formal treatment in anticipation of the Madrid congress.[14]

When the first of these reports appeared, Cochrane[15] had one actually in press describing "intermediate" cases to which the term borderline would have been equally applicable, distinguishing them from major tuberculoid cases. He was evidently dubious about "intermediate" as a class name, however, for in a classification scheme he proposed in 1946[16] one type was called "uncharacteristic or borderline;" it comprised four varieties: *(a)* atypical tuberculoid, *(b)* true intermediate forms, (*c*) sarcoidal, and (*d*) atypical leproma [sic].

A year or so later, at the first All-India Leprosy Conference, Cochrane[17] presented another, simplified, classification scheme without any such complicating class. However, he mentioned another group, "variously known as intermediate or border line cases or atypical lesions" which did not fit into any of the proposed categories, and in the general discussion[17a] he proposed they be made a new type, to be called "atypical."

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[8] WADE, H. W. AND LOWE, J. The type-distribution of patients at the Purulia Leprosy Colony. Indian Med. Gaz. **71** (1936) 653-659; *also* Leprosy in India **9** (1937) 39-48.

[9] It was the examination of these cases and of biopsy material from some of them which particularly aroused the writer's interest in the borderline condition.

[10] LOWE, J. A note on the classification of cases of leprosy. Leprosy in India **10** (1938) 3-6.

[11] LOWE, J. A note on racial variations in leprosy with particular reference to Indian and Burmese races. Leprosy in India **10** (1938) 132-139.

[12] WADE, H. W. AND RODRIGUEZ, J. N. Borderline tuberculoid leprosy. Internat. J. Leprosy **8** (1940) 307-332.

[13] WALE, H. W. Relapsed and borderline cases of tuberculoid leprosy. Leprosy Rev. **12** (1941) 3-17.

[14] WADE, H. W. The classification of leprosy; a proposed synthesis based primarily on the Rio de Janeiro-Havana system. Internat. J. Leprosy **20** (1952) 429-462.

[15] COCHRANE, R. G. Development of the lesions of leprosy, with particular reference to tuberculoid leprosy and the significance of the lepromin test. Internat. J. Leprosy **8** (1940) 445-456.

[16] COCHRANE, R. G. The classification of leprosy. Leprosy Rev. **18** (1946) 36-42.

[17] COCHRANE, R. G. Classification of leprosy. Leprosy in India **20** (1948) 88-91. (*a*) Ibid, p. 35 (discussion).

[18] COCHRANE, R. G. Some brief comments on the classification of leprosy. Leprosy in India **21** (1949) 86-90.

At the next All-India Conference Cochrane,[18] trying "to get away from the stereotyped approach," offered still another new scheme comprising four (actually five) types, one of which was called "dimorphous." Khanolkar had suggested that term (sometimes used in botany and zoology, e.g., "sexually dimorphic") because the previously used ones (intermediate, doubtful, borderline, transitional) were all unsatisfactory. The *type* ("lepra dimorphosus") was described as showing, clinically and histologically, characteristics "of the leprides as well as of leproma;" the *lesions* were spoken of as having "succulent somewhat ill-defined edges" and as being "papulosquamous, squamous, nodular, ulcerative, etc."

The new term did not appear, but the one used in 1940, in the quite different three-type classification scheme which he advanced in 1951.[19] Besides the tuberculoid and lepromatous type there was an "intermediate or atypical" one, which had two subgroups, "atypical lepride" and "atypical leproma."

"Dimorphous" reappeared in a paper presented by Khanolkar and Cochrane at the Madrid congress.[20] Cochrane's three types of 1951 were retained, but the third one was renamed: "borderline (dimorphous)." It now had four subdivisions, the new ones called dimorphous macular and [dimorphous] polyneuritic. With this new application, dimorphous no longer applied specifically to the conspicuous "T?L" cases it originally had. Cochrane used the same scheme in a criticism of the Madrid classification.[21] To bring this account up to date, articles have appeared which deal with the dimorphous macular lesion [22] and dimorphous polyneuritic leprosy [23]—the latter based on examinations of cutaneous nerves, not in conformity with the application of "polyneuritic" exclusively to peripheral nerve trunks customary since the Cairo congress.[24]

In the meantime the original idea of a borderline group gained recognition. The Third Pan-American Conference, held in 1951,[25] pointed out that the reactional form of the tuberculoid type should be well understood because, for one thing, it includes cases presenting features transitional toward the lepromatous form, these constituting the borderline (*limitrofes*) lesions. The WHO Expert Committee, in

[19] COCHRANE, R. G. Classification of leprosy. Mem. III Conf. Panamericana Leprol., Buenos Aires, 1951; Buenos Aires, Vol. 1, 1953, pp. 66-74.

[20] KHANOLKAR, V. R. AND COCHRANE, R. G. Classification of leprosy, with special reference to macules. Mem. VI Congr. Internac. Leprol., Madrid, 1953; Madrid, 1954, pp. 1279-1283.

[21] COCHRANE, R. G. A critical appraisal of the Madrid classification of leprosy. La Lepro **24** (1955) 241-246 (in English); *also* Leprosy in India **27** (1955) 234-240.

[22] KHANOLKAR, V. R. AND COCHRANE, R. G. The dimorphous macular lesion in leprosy. Indian J. Med. Sci. **10** (1956) 499-505.

[23] COCHRANE, R. G. AND KHANOLKAR, V. R. Dimorphous polyneuritic leprosy. Indian J. Med. Sci. **12** (1958) 1-9.

[24] [CAIRO CONGRESS.] The classification of leprosy. Internat. J. Leprosy **6** (1938) 389-397.

[25] [THIRD PAN-AMERICAN CONFERENCE] Clasificatión de subtipos. Mem. Tercera Conf. Panamericana Leprol., Buenos Aires, 1951. Buenos Aires, Vol. 2, 1954, 289-303.

1952, definitely adopted a fourth form of leprosy, borderline.[26] The Madrid congress, in 1953, adopted that form but—reflecting the views of certain members of the classification committee—with an alternative name, dimorphous, in parentheses.[27] The nature of the cases for which the "borderline (dimorphous)" group was established—definitely not macular—is amply indicated by the following quotation from the Madrid report:

> "Such cases may arise from the tuberculoid type as a result of repeated reactions, and sometimes they evolve to the lepromatous type . . . The skin lesions are usually seen as plaques, bands, nodules, etc. . . . The ear lobes are likely to present the appearance of lepromatous infiltration. The lesions frequently have a soft or succulent appearance, and peripherally they slope away from the centre . . . [and] are therefore liable to be mistaken for lepromas."

On this basis it has been perfectly legitimate to speak of these cases in a single breath as "borderline or dimorphous," the two terms synonymous. However, as has been seen, since that group was recognized and defined the term "dimorphous" has repeatedly been applied to lesions of radically different nature. Consequently, there is now uncertainty as to meaning when it is used without adequate qualification.

It seems indisputable that when "dimorphous" is applied to simple flat macular *lesions* (to say nothing of subcutaneous nerves) it is used in a strictly histologic sense—for which *morphe* (form, or shape) is hardly appropriate. It may be expected that leprologists who see the kind of dimorphous macular *cases* described in the report in this issue can learn to recognize them clinically, with such aids as the man in the field has available. We hope to have, in due course, a study of the histopathology of the lesions for our pages.

If a "dimorphous macular" clinical form comes to be accepted, the question will arise as to its proper place in formal classification. The answer is not apparent—unless the present four-class scheme is abandoned in favor of the revolutionary three-type one of Cochrane and Khanolkar. As for differential class diagnosis, the main problem will evidently be to distinguish the indeterminate and dimorphous macules. The lepromatous macular case on the one hand, and what many call the maculoanesthetic case on the other hand, are reasonably distinctive.

Be all that as it may, with the establishment of a dimorphous macular form it will no longer be permissible to speak of "dimorphous leprosy" in the sense of the conspicuously different borderline form. In other words, "dimorphous" can no longer hold place as a parentheti-

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[26] [World Health Organization] Expert Committee on Leprosy; First Report. Wld. Hlth. Org. Tech. Rep. Ser. No. 71, 1953, pp. 19-22.

[27] [Madrid Congress] Technical resolutions. Classification of leprosy. Internat. J. Leprosy **21** (1953) 504-516; Mem. VI Congr. Internac. Leprol., Madrid, 1953; Madrid, 1954, pp. 75-86.

cal alternative for "borderline," and the latter term should regain the specific meaning in classification which—by definition, of course—it originally had.

H. W. WADE