the one as in the other. The pelvis should be carefully estimated in both, and all breech presentations should be delivered in hospital under responsible supervision.

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LABIAL DIPHThERIA

REPORT ON 46 CASES AMONG BRITISH PRISONERS OF WAR IN THE FAR EAST

BY

GEORGE S. RIDDELL, M.D., D.P.H.

Assistant Medical Officer, Aberdeenshire County Council

Labial diphtheria is something of a curiosity, and I confess to having obtained a "literature" only after a fairly extensive examination of British and American publications. How often the lips became implicated in pre-antitoxin days is difficult to assess, though Bretonneau (1821) refers to his colleague Dr. Guersant, physician to the Hospital for Children, as having "frequently observed the pellicular affection of the vermilion part of the lips." Certainly, with the advent of diphtheria antitoxin as a therapeutic agent a labial localization became rare. As instance, Goodall (1928) reports that he "has twice seen membrane on the tongue, three times on the lips, twice on the floor of the mouth, and once on the gums." Top (1941) records only one case with labial membrane in his series of 6,011 patients with diphtheria, while Rolleston (1925) seemingly did not observe such a localization among 3,000 cases of diphtheria.

Rundle (1929) allows that buccal lesions may very rarely be the first sign of diphtheria, but points out that involvement of the buccal mucous membrane, the tongue, and the lips most commonly arises from failure to arrest the spread of the membrane in late severe faucial diphtheria. Cases of this latter order are probably more common than the literature would suggest, for Reiche (1914) records 49 examples of lesions of the lips among 7,314 fatal cases of diphtheria in Hamburg. Instances in which labial membrane has occurred as a separate and distinct localization are certainly much less frequent, although Anderson (1943) has reported a labio-faucial infection and van der Molen (1935) a naso-labial case. The last-mentioned patient, a boy of 3 years, presented membrane on both upper and lower lips, on the tip of the tongue, and in the nostrils. The throat was normal and cultures of the lips yielded diphtheria bacilli.

The most infrequent event of all has been the development of labial membrane unassociated with involvement of either the nose or the throat. Indeed, two American observers, Lavender and Squires (1938), meeting a case of pure labial diphtheria unassociated with involvement of nose or throat in a coloured girl aged 10 years, and believing theirs to be the first recorded instance of this localization, assert that in the literature no mention can be found of cases in which diphtheria involved the lips and buccal membrane alone, with repeated negative clinical signs in the nose and throat. However, a prior claim certainly exists, as

represented by the striking case instanced by Tomb (1925) of diphtheritic stomatitis with membrane on the tongue, lips, and cheek but not on the fauces.

These cases, while in no way purporting to represent a complete survey of the literature of labial diphtheria, suffice to emphasize the rarity of this localization and to allow the conclusion that membrane may appear on the lips in the following circumstances: (1) as a direct extension of membrane in late severe faucial diphtheria; (2) as a very occasional accompaniment of nasal or faucial diphtheria; and (3) as an exceptional phenomenon in the absence of diphtheritic infection of the nose and throat.

The Cases

This paper relates to a group of 46 cases of labial diphtheria observed among British prisoners of war in a Japanese prison camp on Singapore Island during the period February 15, 1942, to February 15, 1943, and constituting part of an extensive epidemic of 940 cases of diphtheria, both respiratory and non-respiratory. In 24 instances no focus of diphtheria other than the lips seemed to exist. In four of the early examples negative cultures were obtained from the nose and throat. Thereafter shortage of material and pressure of work on our limited laboratory facilities, already fully extended, compelled us to refrain from taking such cultures provided the throat and nose were clinically clear. An associated localization was present in 22 patients, the second focus being an anterior nasal rhinitis in three, faucial diphtheria in eight, naso-faucial diphtheria in four, and a coexisting cutaneous diphtheritic lesion in seven.

The Predisposing Lesion

The key to the occurrence of the labial affection lay, beyond all question, in the existence in every case of a definite, specific predisposing lesion. This lesion, which was present for days or even weeks before the advent of the diphtheritic disease, comprised the highly characteristic cheilosis, maceration, and fissuring at the corners of the mouth first brought to the notice of the medical profession as a probable deficiency state by Stannus (1912, 1913) under the name "angular stomatitis" and finally differentiated as an expression of riboflavin deficiency by Sebrell and Butler (1938). The labial manifestations of hypo-riboflavinosis were of extreme frequency and often considerable severity among the prisoners. Severe cases not infrequently showed some heaping up of sodden, macerated, whitish epithelium at the corners of the mouth, but this bore no real resemblance to a true labial membrane and did not give rise to confusion.

Slight trauma to the lip in the presence of an existing faucial infection is considered by Anderson (1943) to be the predisposing factor in his case of labio-faucial diphtheria, and he expresses surprise that labial membrane is not more often seen. In view of the apparent readiness with which lesions of the lips became infected in my series and how widespread are minor lesions of the lips—minor trauma, impetigo, herpes labialis, and the like—in normal practice, it is indeed remarkable that labial membrane has long remained a curiosity.

Bacteriology

In the circumstances prevailing in a Japanese prisoner-of-war camp complete bacteriological finality was not possible. Single cultures for the *Corynebacterium diphtheriae* were taken from the lips in 25 cases, and 19 of these proved

positive at this first attempt. In four cases with a coexisting respiratory localization and in four with a concomitant cutaneous lesion cultures were also taken from the second focus of infection, in each case with a positive result. Diagnosis otherwise rested on clinical grounds alone, but the associated circumstances of the epidemic, the characteristic labial lesion, and the isolation of the specific organism in 19 cases provide ample evidence that the cases were instances of true diphtheritic membrane formation on the lips. Virulence tests were beyond our resources.

The Clinical Picture

The advent of diphtheria on the deficiency manifestation gave rise to a distinctive, almost pearly white or faintly yellowish, raised, glistening membrane, firmly adherent and covering as a rule the outer two-thirds of the upper and lower lips on each side, but not infrequently extending to meet in the midline. The membrane was always most pronounced at the corners of the mouth, but only very exceptionally was it unilateral. So striking was the clinical picture that the diagnosis rarely, if ever, gave rise to any difficulty to unit medical officers even when faced with a case for the first time. The existence of a second focus of infection was without apparent influence on the appearance of the labial localization.

Toxaemia was absent or quite insignificant in the purely labial case. Cervical and submental adenitis of slight degree was present in most instances. In the absence of therapeutic antitoxin the membrane separated in some seven to ten days to leave an area of very shallow and superficial ulceration which healed readily without scarring. Where serum was given in a purely labial case (two instances) the membrane separated within 48 hours.

In the group of cases with a double infection it was hoped that careful histories might distinguish the primary localization. In the three naso-labial cases the answer was definite, all three patients providing a clear history of nasal obstruction and discharge for several days before the appearance of membrane on the lips. In the seven cutaneous and labial cases it was again quite evident that the labial localization was a secondary phenomenon doubtless due to transfer of infection through the medium of the hands. By contrast, in at least seven of the twelve labio-faucial cases it seemed possible if not probable that the lips had been the primary focus, this leap from lips to fauces constituting the great, indeed the only, danger of the labial infection. However, no such deterioration occurred in a case of labial diphtheria under my observation. It must be emphasized that in all these cases the two lesions were anatomically separate and not due to direct extension of the membrane from one area to the other.

End-results : Complications

No fatalities occurred in the group of 24 purely labial cases, nor did the slightest indication of post-diphtheritic neuritis or clinical myocarditis appear in any instance. This localization of diphtheria *per se* would therefore seem to be extremely benign, extremely characteristic, and dependent on a suitable predisposing lesion—in my cases a highly specific one. The only danger inherent in labial diphtheria, and that a very real one, was the risk of spread of infection to the fauces as discussed above.

The outcome of events in cases with a double infection was governed by the major localization. The three naso-labial cases had no fatalities, sequelae, or complications, and all seven cases with labial membrane and cutaneous

diphtherial involvement made uneventful recoveries. The twelve labio-faucial cases had no fatalities, but clinical evidence of myocarditis (persistent severe tachycardia) appeared in two instances, both of which subsequently developed neuritis—in one paraesthesia and absent reflexes; in the other hypalgesia, impaired vibration sense, and mild peripheral motor neuritis in the lower extremities. Neuritic phenomena appeared in four other cases—paralysis of accommodation in one, palatal paralysis in two, and palatal palsy plus peripheral motor neuritis in one. Neurological involvement thus complicated no fewer than six of the twelve labio-faucial cases.

Treatment

The usual regimen of strict rest applicable to cases of diphtheria was carried out so far as lay in our power. On the whole this aspect of the management of the cases could be regarded as satisfactory and probably accounts for the low incidence of complications despite the lack of antitoxin. Therapeutic antitoxin was available for five cases only—two purely labial and three labio-faucial. Of the latter, two escaped without complications and one developed post-diphtheritic neuritis.

Summary

Certain aspects of the somewhat limited literature of labial diphtheria have been reviewed, and an account is given of 46 cases with labial membrane among British prisoners of war in the Far East. In 24 of these no focus of diphtheria other than the lips seemed to exist; in 15 there was an associated respiratory localization; and in 7 a coexisting cutaneous focus of infection. The labial cases were part of an extensive outbreak of diphtheria.

The infection of the lips depended on a specific predisposing lesion—fissuring at the angles of the mouth and cheilosis of the vermilion part of the lips—following deficiency of riboflavin in the diet.

Labial diphtheria alone or associated with anterior nasal or cutaneous diphtheria was unattended by mortality, complications, or sequelae. The presence of a faucial infection brought with it the dangers and complications of that localization.

Since purely labial diphtheria was unassociated with either myocarditis or neuritis, as were the seven cases in which the lips and the skin were implicated in the absence of clinical signs in the nose and/or throat, the cases failed to provide an opportunity of observing the cardiac and neuritic complications of diphtheria as seen in non-respiratory localizations, and are chiefly of interest in that they provide a large series of a distinctly rare localization.

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