CASE NOTES

UNUSUAL CASE OF INTERSTITIAL KERATITIS*

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THE clinical features of interstitial keratitis—a fast disappearing disease—are well known and so definite as to enable not only ophthalmologists but clinicians in general to arrive at a correct topographical if not aetiological diagnosis. That there are atypical forms of almost all diseases is indisputable and the following case report appears justifiable because of its close mimicry of Mooren's ulcer, and its defiance of all forms of treatment including an annular lamellar keratoplasty.

Case Report

A married woman aged 23 first reported with a painful red eye of a week's duration on November 14, 1954. She had had no previous ocular trouble but was reticent about previous venereal illness.

Examination.—The appearances at the time of first examination were those of a marginal ulcer of the cornea commencing at the inferior limbus of the right cornea. Within a week of commencing treatment the marginal "ulcer" spread all round the limbus and centripetally, undermining the cornea and producing overhanging edges. An extremely vascular pannus crept stealthily under the superficial layer of the cornea, segregating an island of superficial grey-looking cornea precariously perched on the deeper corneal substance. The "ulcer" also spread towards the episcleral tissue in the neighbourhood of the limbus. The whole picture presented a sinister appearance, reminiscent of Mooren's ulcer. The visual acuity at this stage was 6/60 in the right eye and 6/9 in the left (Fig. 1 *a* and *b*).

FIG. 1 (*a*).—Right eye showing corneal "ulcer" with overhanging edges typical of Mooren's ulcer.

FIG. 1 (*b*).—Key to Fig. 1 (*a*).

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At the time of hospitalization, a week after she was first seen in the clinic, the left eye too showed a marginal "ulcer" at the temporal limbus and the visual acuity had dropped to 6/12.

Biomicroscopic examination revealed oedema of the superficial cornea, overhanging edges at the margins of the "ulcer", and a rich pannus working its way underneath the corneal substance as it extended leaving in its wake a vascularized scar. The pannus did not extend beyond the edge of the ulcer. At no stage of the active disease was it possible to examine the iris, though at the commencement and after the resolution of the inflammation the iris was normal and there were neither residual stigmata of iritis nor changes in Descemet's membrane.

Laboratory Investigations.—Blood Wassermann reaction negative on November 25, 1954. X ray of chest normal.

Treatment

(a) *Medical.*—Streptomycin drops 4-hrly and atropine drops twice daily were instilled. In hospital topical cortisone 1 per cent. ointment was given a trial for 2 weeks without benefit. Riboflavin in large doses was tried by mouth with equivocal results. On December 27, 1954, a course of streptomycin (total 14 g.) and iso-nicotinic acid hydrazide (total 1,400 mg.) was started, in the hope of attacking a probable tuberculous aetiology. In February, 1955, the right eye had reached a hopeless state (visual acuity 1/60) and the left eye was also deteriorating (visual acuity 6/18). A further course of a combined preparation of dihydrostreptomycin and tri-isonicotinyl hydrozone pyruvate (Streptotibine, Lepetit) was started, 1 g. daily being given intra-muscularly for 10 days. On February 8, 1955, the patient was admitted to hospital for a keratoplasty on the right eye.

(b) *Surgical.*—An annular lamellar grafting was performed as follows: two cuts were made with 10- and 6-mm. Franceschetti trephines so as to obtain a signet-ring-shaped graft (Fig. 2). This was necessary as the "ulcer" had spread more inferiorly than elsewhere. The graft was cut with a Bock's knife in steps by rotating the donor eye by small amounts. As the central 6 mm. disc of donor cornea was required for another patient one had to be careful in shaping the annular graft.

FIG. 2.—Outline of (a) limbus and site of 10 mm. trephine cut; (b) "ulcer" edge; (c) 6-mm. trephine cut; (d) pupil. Note signet-ring-shaped area of keratectomy and graft.

The recipient (right) eye was prepared by employing 6- and 10-mm. trephines. To trephine the recipient eye, especially along the overhanging edges of the ulcer, was extremely difficult owing to the unsteadiness of the undermined fringe of oedematous and friable cornea. The area between the two trephine cuts was dissected with a Desmarres' keratoplasty knife and the bleeding points touched with hot probes. The graft was transferred to the bed and secured by overlay sutures over egg membrane.

Progress.—The grafted eye had a stormy course, but with antibiotics and cortisone topically the reaction abated. The graft had taken but was overrun by a girdle of blood vessels which quickly engulfed it; 10 days after the grafting the ulceration had progressed more centripetally (Fig. 3 a and b, opposite). The left eye was now passing through a phase of creeping ulceration similar to that in the right eye. Owing to domestic difficulties the patient left hospital but continued to attend the clinic regularly.

In May, 1955, the visual acuity had fallen to hand movements in the right eye and 1/60 in the left. On our suggestion she re-entered hospital on May 24, 1955 for a second

FIG. 3 (a).—Same eye after keratoplasty. "Ulcer" edge has encroached on cornea, and peripheral parts of cornea are scarred.

FIG. 3 (b).—Diamond-shaped island of intact cornea; two edges below it bound a tongue-shaped area where the graft has failed to take.

graft operation on the right eye. When she was admitted an eruption on the lower abdominal skin and groin and a vaginal discharge were discovered. The venereologist reported that she had been attending his clinic for some time. Antisyphilitic treatment was therefore recommenced and the idea of a second operation was abandoned, partly because of the skin condition and partly because the aetiological agent was now disclosed. The corneae started clearing after 2 months and the patient was able to attend the clinic without a guide. In August, 1955, the visual acuity in the right eye was 6/36, and in the left 6/60; the corneae were clearing, the pannus had thinned appreciably, and there were no permanent changes in the iris or Descemet's membrane.

Discussion

The vagaries of interstitial keratitis of whatever cause are well known. In both the syphilitic and tuberculous types the deeper layers of the cornea are involved. Clearing of the cornea is more thorough in the former, whereas permanent dense white scars appear to be the rule in the latter. The epithelium is not affected in either condition, and in the tuberculous type abscess-like infiltrates are sometimes seen (Duke-Elder, 1937).

But for the age of the patient, a diagnosis of Mooren's ulcer was the only one possible, and this was our tentative diagnosis qualified by the prefix "atypical". The early clinical appearances of a grey marginal infiltrate of the cornea soon spreading along the limbus, and excavating the corneal substance, a pannus creeping centripetally, and a thickening of the episcleral tissue in the immediate vicinity of the limbus, are too typical for such a diagnosis to escape one's mind.

The occurrence of interstitial keratitis in a sero-negative person (in this instance due to previous antisyphilitic treatment) is not unusual; the corneal condition in this case was not a tuberculous ulceration of the cornea as there was no primary tuberculosis of the lids or conjunctiva. The question of a silent tuberculous focus was considered, because we could find no other incriminating cause.

In the absence of a definite aetiological factor and owing to the rapid deterioration of sight, we felt it rational to perform a keratoplasty. An annular lamellar keratoplasty was chosen because a central fairly normal cornea was still present and because we thought it would eliminate the halo of pannus undermining the cornea and bridge the gap between the limbus and the normal centre. The blood vessels threatening to invade the cornea were not checked by the annulus of fresh corneal tissue, however, because of the overwhelming nature of the pathological process.

A lesson to be drawn from this case is that in every instance of Mooren's ulcer a thorough search must be made for syphilis and tuberculosis, for this condition may be an atypical form of syphilitic or tuberculous interstitial keratitis (Koepe, 1918).

Conversely, there was nothing definite in this case to indicate that it was in fact due to interstitial keratitis. It may have been a case of Mooren's ulcer, atypical only on account of the youth of the patient—for Taylor has described this condition in a child aged 3 (Linn, 1949). The undermined edges of the ulcer, a creeping pannus throwing a veil over the cornea as it extended, bilaterality, chronicity, and resistance to antibiotics and cortisone, are typical of Mooren's ulcer, as are the absence of changes in the iris and Descemet's membrane. However, the recovery of corneal clarity after such devastating changes is rare in Mooren's ulcer, and rather resembles the pattern of events in interstitial keratitis. For this reason the patient's improvement can only be attributed to the extensive antituberculous and antisyphilitic measures adopted.

It is suggested that this case should be classed as an atypical form of interstitial keratitis with a mixed tuberculous and syphilitic aetiology, and that Mooren's ulcer should be regarded as a form of interstitial keratitis and not as a separate clinical entity. It also seems appropriate that the condition termed Mooren's ulcer should be called Mooren's syndrome since there may be more than one cause for the classical clinical picture of a "chronic superficial ulcer . . . which with a characteristically undermined border and without perforation progresses slowly and unrelentingly . . . until the whole cornea may be involved" (Duke-Elder, 1937).

The efficacy of therapy will depend on the aetiology. Corkey (1952) successfully treated a case of Mooren's ulcer with riboflavin, but in this case we did not meet with the same success, nor did keratoplasty have any appreciable effect.

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