INTERMITTENT MELANOPHORA*

BY

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A 14-year-old girl suffered from an intermittent discharge into the conjunctival sac of a black sticky material, which was washed out by tears and could occasionally be removed by a cotton swab. Neither we nor Prof. Ida Mann, who saw the patient in October, 1962, had seen or heard of such a condition before, and no such description could be found in the literature.

Case Report

A young Muslim girl aged 14 years (Fig. 1), resident in Aligarh, came to the out-patients department of the Gandhi Eye Hospital on June 14, 1962, complaining of the occasional appearance of a blackish pigment in the left eye for the past month (Fig. 2).

FIG. 1.—The patient.

FIG. 2.—Collection of free sticky melanin pigment in left lower fornix. Note identical impressions of palpebral pigment on bulbar conjunctiva, July, 1962.

History.—While she was working in a science laboratory in January, 1962, some chemical had fallen into her left eye and injured it. The pigment first appeared in the lower fornix in May, 1962.

It was suspected at first (because she had access to chemicals) that she might herself be putting some substance into her eye. The pigment was cleaned out every day and antibiotic ointment was applied, and in the meantime her family background was investigated, but nothing significant was discovered.

Treatment.—On July 3, 1962, she was admitted to hospital for observation; after the eye had been cleaned, antibiotic ointment was applied, and the eye was padded, sealed with Elastoplast, and bandaged. A nurse was posted to watch the patient, but 48 hours later the sticky pigment was found to have collected in the left lower fornix. The eye was again cleaned and bandaged and a strict watch was kept, but after 24 hours a fresh collection of pigment was found.

There was no sign that the bandage had been tampered with, but we were still unconvinced; we therefore repeated the process yet again, and searched her belongings to see if any chemical could be found, but there was no clue whatever to support the idea that she was malingering.

We therefore searched for some organic lesion, but all our local and general investigations gave negative results.

The sticky material, which was found to consist of melanin pigment, first appeared as a blackish-brown subconjunctival spot and gradually grew in size like a mass of black paste in the lower fornix, some of which could not be removed without injuring the conjunctiva. The activity was

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intermittent and irregular. It would increase every day for some days and would then quieten down for some days and sometimes even for weeks. The pigment was produced mostly on the tarsal conjunctiva near the fornix and would also adhere to the bulbar conjunctiva.

Investigations.—Stools, urine, and blood were normal. The Kahn and Mantoux tests were negative. A smear showed only scattered pigment granules. No fungus could be cultured. The pigment could be decolorized by potassium chlorate and concentrated hydrochloric acid and also by potassium permanganate and oxalic acid. Tests confirmed that it was in fact melanin.

Treatment.—The area was cauterized with iodine followed by beta-irradiation, but the pigment reappeared after 14 days. It was therefore decided on August 4 to excise the area of the lower fornix with a partial tarsectomy and mucous membrane graft. Recovery was uneventful and the stitches were removed on August 13 (Fig. 3). Local antibiotics and oral colloidal iodine were given.

Histological Report.—In the section from the conjunctiva and tarsus, the epithelium was thinned in places and showed hydropic degeneration. The subepithelial tissue showed chronic inflammatory reaction with evidence of acute exacerbation. There was a deposit of melanin-like pigment in the basal layers of the conjunctival epithelium, and in the subepithelial tissues.

Follow-up.—To our surprise, the patient returned on August 25 with a little pigment in the left lower fornix, the starting point of further pigment activity below the mucous membrane (Fig. 4).

The pigment has continued to collect periodically in the left lower fornix and the patient removes it herself or occasionally comes to the hospital (Fig. 5). In December, 1962, the disease started in the right lower fornix also. She has been advised to have low-dosage x rays, but for fear of cataract prefers to put up with her present inconvenience. She has developed slight scarring in the left lower fornix with a small posterior partial symblepharon (Fig. 6). It seems that the pigment activity comes in cycles but has no connexion with the normal menstrual cycle. She was last examined on August 24, 1963.

FIG. 3.—Left lower fornix on August 13, 1962. Conjunctiva, subconjunctival tissues, and part of the tarsus were excised and a buccal mucous membrane grafted on.

FIG. 4.—Reappearance of pigment on August 25, 1962, after mucous membrane graft.

FIG. 5.—Appearance in December, 1962.

FIG. 6.—Appearance on August 24, 1963. There had been no pigment activity for 12 days. Only a posterior partial symblepharon is seen after 16 months of treatment and recurrence.

Discussion

In this case, although the histopathological picture is consistent with conjunctival melanosis, the pigment forms in the basal conjunctival cells, and instead of spreading sub- or intra-epithelially discharges freely as a paste in the lower fornices. It can be cleaned with a swab or is washed away by tears, but part remains embedded sub-conjunctivally. Occasionally it disappears completely.

According to Reese (1938, 1943) the earliest changes in melanosis are seen in the basal epithelial cells which become highly pigmented and hydropic and suffer from globular distension. At places the basal cells proliferate, aggregate into clumps, and invade the subepithelial layers of the conjunctiva, or infiltrate the normal conjunctival structure. Duke-Elder (1952) also mentions it as mainly intracellular, in the basal cells, epithelial cells, or chromatophores.

In this case there appears to be a localized excessive production of melanin in the basal layers of the conjunctiva so that the cells burst and liberate the melanin into the fornix. Such excessive melanin production may be due to the action of tyrosinase or a similar compound on hydroxy phenyl alanine (Krause, 1933). The periodical benign increased production of melanin may have something to do with the endocrine glands, particularly the pituitaries or suprarenals, but no endocrine disturbance was found in this case. The continued pigmentary production after the mucous membrane graft may be due to the activation of migrant basal conjunctival cells which were incompletely excised.

Summary

A case of localized intermittent melanin production in the lower fornices of both eyes is reported for the first time. The free melanin was discharged into the conjunctival sac and could be wiped away or washed away with tears.

REFERENCES

- DUKE-ELDER, S. (1952). "Text-book of Ophthalmology", vol. 5, p. 5108, Kimpton, London.
 KRAUSE, A. C. (1933). *Arch. Ophthalm. (Chicago)*, **10**, 42.
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 ——— (1943). *Ibid.*, **29**, 737.

ADDENDUM

Since this paper was submitted, the patient has been given fortnightly intramuscular injections of whole pituitary extract, and has had no recurrence during the past 8 months, except once in the right eye in January, 1964, and that was very mild and spotty and lasted only about a week. This proves that the pituitary has a definite role in melanin production. In our case the deficiency of a certain factor may have released the inhibition on potential melanin-producing cells. The basal conjunctival cells, particularly those at the limbus, are prone to melanin production as is evident from the wide and variable pigmentation of that region in both animals and man. The administration of pituitary extract may prove useful in treating various types of melanotic disturbance.

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