Primary orbital aspergilloma

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Aspergillosis of the orbit producing proptosis is a rare condition, only thirty authenticated cases having been reported in the literature up to September, 1969.* Emmons, Binford, and Utz (1963) referred to another ten cases involving the paranasal sinuses or the orbit or both, in which the characteristic feature was chronic densely-fibrosed granulomatous tissue. In most of these cases, the infection apparently began in one of the accessory nasal sinuses and the orbital involvement was secondary.

Fungus infection of the orbit may be granulomatous or non-granulomatous, and may lead to the development of an abscess. The granulomatous type usually manifests as proptosis with limitation of movement and the non-granulomatous type may give no sign apart from an infected sinus. Some cases of fungus granuloma of the orbit may be misdiagnosed as pseudotumour unless special staining techniques are used. The following case observed in northern India showed clinical and histological features which differed from those hitherto reported.

Case report

A 40-year-old housewife was admitted to hospital on December 15, 1962, with progressive and painful protrusion of the right eye over a period of 3 months. There was no history of trauma, cough, sinusitis, fever, or diplopia, and systemic and E.N.T. examinations revealed nothing abnormal. There was no enlargement of the preauricular nodes.

Ophthalmological examination

The right eye was displaced forwards and slightly upwards, the proptosis of 5 mm. being non-pulsatile and non-reducible (Fig. 1). No mass could be seen or palpated in the orbit. Movements of the eyeball were normal. The lens showed cataractous changes. Visual acuity was 5/60 and J4. Fundus examination showed slight hyperaemia and blurring of the margins of the disc and engorged veins.

FIG. 1 *Right proptosis*

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* Dalmeijer (1913), Wright (1927), Adams (1933), Nash (1938), Tikhomirov (1958), Weller, Joseph, and Hora (1960), Bailey and Fulmer (1961), Agarwal, Malik, Mohan, and Mahopatra (1962), Seabury and Samuels (1963), Agarwal, Dayal, and Agarwal (1964), Satyendran, Reddy, and Krishnamoorthy (1964), Miloshev, Davidson, Gentles, and Sandison (1966), Green, Font, and Zimmerman (1969).

Laboratory investigations

The basal metabolic rate, X-ray films of the chest, skull, paranasal sinuses, and orbits, blood studies, urine analysis, and Mantoux and Casoni tests were of no significance. The erythrocyte sedimentation rate was 29 mm./1st hour (Westergren). A smear from the conjunctival sac was negative on culture.

Treatment

In view of the relatively rapid progress of the condition and the negative results of these investigations, the possibility of an orbital pseudotumour was considered and oral steroids and systemic antibiotics were given but without any benefit. An anterior orbitotomy was performed 4 weeks later under general anaesthesia. A greyish-white, irregular, firm and circumscribed mass measuring $2 \times 1.5 \times 0.5$ cm. was excised. A gritty sensation was felt on sectioning. The mass extended along the floor of the orbit towards the apex.

Microscopic examination

Haematoxylin and eosin stained sections showed an abundant chronic inflammatory exudate rich in epithelioid cells, giant cells mostly of the foreign body type, some plasma cells, lymphocytes, and histiocytes. There was incipient tubercle formation. No area of caseation was seen. The tissue was moderately vascular.

Progress

Antitubercular therapy was instituted because of the histological findings, but there was no improvement and the pathological specimen was reviewed. On closer scrutiny, a few broad-segmented septate hyphae of variable lengths were seen both within the giant cells and outside them (Fig. 2, opposite). This was consistent with the diagnosis of aspergilloma. Gram's stain and Ziehl Neelsen stain showed no bacteria, but Gridley's stain showed numerous hyphae (Fig. 3, opposite).

Since the tissue had already been fixed in formalin, no further growth of the fungus was possible. No medical anti-fungal therapy could be given as the patient did not return to the clinic.

Comment

Among 28,927 patients who attended the ophthalmic out-patients department in 1962, fourteen cases of non-endocrinal proptosis were recorded, of which one proved to be a case of aspergilloma of the orbit. Of the total series of thirty cases so far recorded, six have been reported from India (Wright (1927) two; Agarwal and others (1964) three; Satyendran and others (1964) one). The species of *Aspergillus* producing a granulomatous orbital mass included *fumigatus* (1), *oryzae* (2), *flavus* (6), *terreus* (1), and *flavus-oryzae* (2); in the other eighteen cases no species could be identified. In 77 per cent. of these cases uniocular proptosis was the first sign of the disease, and the next most common sign was involvement of the paranasal sinuses. Systemic involvement was reported only by Nash (1938).

Fungi in the nasopharynx and sinuses may extend into the orbit to produce orbital cellulitis or a mass or may involve the optic nerve sheath to produce optic neuritis. Ocular and orbital extension may be derived from cerebral involvement but trauma has also been present in a few cases. Our patient had not suffered any injury nor was there any clinical or radiological evidence of mycosis in the paranasal sinuses, so that the source of infection remains obscure. As the involvement was restricted to the orbital region, it is presumed that this was a case of primary fungus granuloma of the orbit. It is the fourth case to be reported, the first three having also come from India: Agarwal and others, 1964 (2); Satyendran and others, 1964 (1). The other 27 cases showed concomitant

FIG. 2 Granulomatous process, showing hyphae within the giant cell and outside. Haematoxylin and eosin, $\times 250$

FIG. 3 Specimen stained with Gridley's stain, showing numerous hyphae. $\times 200$

involvement of the sinuses, central nervous system, and lungs. Satyendran's case may not have been one of primary orbital aspergilloma in view of the positive radiology of the sinuses.

The non-reducible nature of the proptosis, the congestion of the retinal veins, and the appearance of the optic disc suggested the presence of a firm mass in the orbit and this was proved by surgical exploration, but the raised erythrocyte sedimentation rate gave the only other clue.

Summary

In a case of unilateral proptosis, which simulated an orbital pseudotumour, the circumscribed mass was removed from the right orbit by anterior orbitotomy. The correct diagnosis was established only by special histological staining for mycosis. It is emphasized that, without special investigations, the diagnosis may be confused with chronic granuloma of tubercular aetiology.

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