Epipalpebral conjunctival osseous choristoma

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SUMMARY Two cases of osseous choristoma are described. One of the tumours was found in the conjunctiva of the right lower lid, an apparently unique location. The other more typical epibulbar tumour was found in the superior temporal quadrant of the episclera between the lateral and superior rectus muscles. In both instances the tumour was suspected of being a dermoid.

Osseous choristomas are rare lesions of the conjunctiva, episclera, and choroid. They usually are described as a pea-to-almond size nodule of compact bone surrounded by a connective tissue capsule. They appear mainly in the superior temporal quadrant of the episclera, between the lateral and superior rectus muscles. There have been at least 38 cases of epibulbar and 5 cases of choroidal osseous choristomas described (von Gräfe, 1863; Spencer-Watson, 1871; Saemisch, 1876; de Wecker and Landolt, 1880; Critchett, 1882; Walker, 1882; Loring, 1883; Snell, 1884; Vignes, 1889; Wagenmann, 1889; Cirincione, 1895; Galtier, 1895; Hartridge, 1895; Nettleship, 1895; Heustis, 1899; Parsons, 1904; Collins, 1927; Ballantyne, 1940; Boniuk and Zimmerman, 1962; Delmotte *et al.*, 1962; Wiesinger and Guerry, 1962; Beckman and Sugar, 1964; Sihota, 1964; Kaufer and Plater, 1968; Roch and Milauskas, 1968; Kreibig and Nehm, 1969; Ferry and Hein, 1970; Fanta, 1977; Gass *et al.*, 1978). None of the previously reported cases involved the lower lid.

Case reports

CASE 1

A 13-year-old boy was noted by his mother to have 'tissue resembling a scar' over the conjunctiva of his right eye. Ophthalmological examination revealed a 5 × 5 mm lesion in the superior temporal quadrant of the right globe between the lateral and superior rectus muscles (Figs. 1 and 2). The remainder of the ocular examination showed no abnormalities. The clinical diagnosis was dermoid, right eye.

Macroscopically the specimen consisted of a hard lenticular fragment of tissue measuring 4 × 3 × 2 mm.

Fig. 1 Case 1. The superior temporal quadrant reveals a raised lesion

Microscopically a fragment of bone surrounded by collagenous connective tissue was noted (Fig. 3).

CASE 2

An 8-month-old girl had a lesion protruding from the temporal side of the conjunctiva of her right lower lid; there was no connection to the bulb. The lesion was first noted at the age of 6 weeks. At that time the child was brought to the ophthalmologist because of increased tearing of the right eye. The clinical diagnosis was dermoid of the right lower lid. Over the next 6 months the child had progressive mucopurulent discharge from both eyes which seemed to be secondary to obstructed nasolacrimal ducts. The lesion in her right lower lid was thought to have enlarged slightly and was removed. At the

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Fig. 2 Case 2. At surgery a $4 \times 3 \times 2$ mm oval-shaped osteoma was excised

time of surgery the lesion was diagnosed as pyogenic granuloma. The dacryostenosis was relieved by probing and irrigation.

Macroscopically the specimen consisted of an $8 \times 3 \times 1$ mm fragment of hard, white, nodular tissue. Microscopically a spicule of bone with haemopoietic marrow was noted (Fig. 4).

Discussion

There have been scattered reports of unusual appearances of osseous choristomas. Roch and

Milauskas (1968) had 2 cases of epibulbar osseous choristoma which extended to the lateral rectus muscle sheath. A similar finding was also described by Ferry and Hein (1970). Fanta (1977) noted an osseous choristoma arising from the outer canthus of the left eye with no connection to the bulb. Gass *et al.* (1978) described 5 cases of choroidal osseous choristomas, 1 of which was confirmed by histopathology. Although also characteristically occurring in young women, the relationship of these intraocular tumours to lesions of the episclera is not clear.

In the 43 reported cases, as in the ones described here, the superior temporal epibulbar location of the osseous choristoma was cited in 23 (51%) of the cases. The temporal epibulbar location was described in 14 (31%) of the cases; the exact site was not listed in 2 (4%). Choroidal osseous choristomas were found in 5 (11%) of the cases. No report could be found where the lower lid was involved.

The majority of epibulbar tumours are discovered and excised by the time the patient is 14 years of age. They have been found to be twice as common in females.

In our Case 1 the lesion was found in the temporal outer quadrant of the episclera in an adolescent. This location is the same as in the majority of cases of epibulbar osseous choristomas. In our Case 2 the lesion was found in the temporal side of the conjunctiva of the right lower lid, in a child with bilateral dacryostenosis. The question is raised here of the possibility that the tumour may have been related to chronic inflammation. Galtier (1895), Boniuk and Zimmerman (1962), and Roch and Milauskas (1968) list several cases in which patients had a history of trauma preceding the appearance

Fig. 3 Case 1. Bone encapsulated in a connective tissue capsule (H and E $\times 10$)

Fig. 4 Case 2. Zones of haemopoietic marrow are seen in the fragment of bone (H and E $\times 12$)

of the osseous choristoma. Boniuk and Zimmerman (1962) speculated that the trauma may have alerted the patient to an already pre-existing lesion. Other causes proposed (in addition to congenital, inflammatory, and traumatic) include atavistic remnant (Boniuk and Zimmerman, 1962; Roch and Milauskas, 1968) and embryonic rest of the mesodermal layer (Roch and Milauskas, 1968). Collins (1927) thought that the lesion could be a result of embryological metaplasia. Because most of the tumours are discovered in infancy and are usually located between the lateral and superior rectus muscles, the congenital cause seems the most plausible.

Epibulbar osseous choristomas are often confused clinically with epibulbar dermoids. Boniuk and Zimmerman (1962) elucidated the essential differences between the two lesions: (1) Epibulbar osseous choristomas are composed entirely of compact bone whereas epibulbar dermoids are composed of associated dermoid structures; (2) epibulbar dermoids are usually located in the inferior temporal rather than the superior temporal quadrant of the globe; and (3) epibulbar dermoids are frequently (30%) associated with congenital anomalies, usually involving malformations of the branchial arch derivatives (Crawford, 1976), whereas epibulbar osseous choristomas are thought to be a result of the above-described causes.

Beckman and Sugar (1964) noted that a simple pre-

operative x-ray examination of a suspected lesion would detail the radio-opacity of the osseous choristoma and help in the differential diagnosis of osseous choristoma from radiolucent dermolipomas and epibulbar dermoids. Two cases of epibulbar dermoids associated with bone have been reported, but they are isolated cases (Wagenmann, 1889; Ferry and Hein, 1970).

In previous reports these bony tumours have been termed epibulbar osteoma (Ballantyne, 1940; Sihota, 1964; Roch and Milauskas, 1968), episcleral osseous choristoma (Boniuk and Zimmerman, 1962), epibulbar osseous choristoma (Beckman and Sugar, 1964; Ferry and Hein, 1970), and choroidal osteoma (Gass *et al.*, 1978). But, as seen in our second case, the tumour can also be found in the conjunctiva of the lower lid. Although it is a minor point, we consider that epipalpebral conjunctival osseous choristoma is a proper term for this variety of osseous choristoma, since it defines the exact location of the lesion and emphasises its non-attachment to the bulb.

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