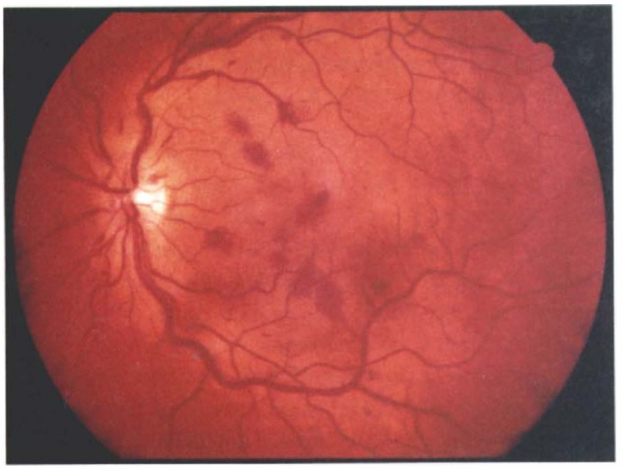
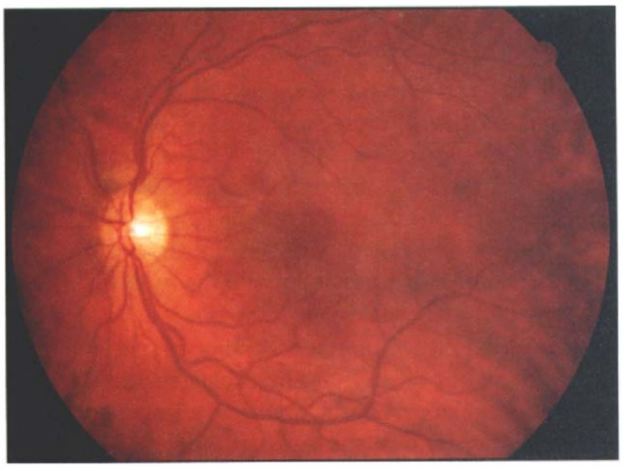


(a)



(b)

Fig. 1. *Left fundus photograph showing retinal haemorrhages at presentation (a) and after 4 weeks (b).*

Valsalva retinopathy following inflation of a party balloon. We believe the rise in the intrathoracic pressure needed to inflate these long balloons produces a significant Valsalva stress. The reason why the retinopathy occurs unilaterally is unknown. A review of 16 cases[1–7] shows no preference for the left or right eye to be affected (7 cases left, 9 cases right).

Although the macular haemorrhages are accompanied by an acute reduction of visual acuity, resolution occurs by 6 months and most resolve within 1–3 months. No specific treatment is recommended but we advise patients to rest and avoid strenuous activity until resolution. Fluorescein angiography is helpful to exclude any other causes of retinal haemorrhage.

A recent report has highlighted the risk of ocular blunt trauma with party balloons exploding during inflation[8] and recommended the use of an inflation device. We support this recommendation to avoid Valsalva retinopathy.

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Sir,

Pedunculated dermolipoma with overlying upper lid coloboma and absent lateral canthus: cause and effect?

Dermolipoma is a subconjunctival choristoma and commonly appears as a soft, yellow, movable subconjunctival mass at the temporal fornix.[1] Coloboma of the upper eyelid is a rare congenital defect and usually involves the medial part of the lid. The two may rarely coexist, usually in the oculofacial syndromes such as Goldenhar's syndrome and epidermal naevus syndrome.[2]

We report here the case of an infant with a pedunculated dermolipoma at the lateral canthus associated with coloboma of the lateral third of the upper lid and absent lateral canthus without associated oculofacial syndrome. This rare combination along with the unusual histology of the tumour prompted us to report the case. We discuss the surgical management of this tumour and propose a hypothesis regarding its possible role in causing the lid coloboma.

Case report

A 6-month-old Omani girl was seen because of a mass protruding from the left eye since birth. Examination showed a pedunculated, soft mass 6 mm × 4 mm in size at the lateral canthus, replacing the canthal angle. The mass was covered on the medial side by thickened conjunctiva changing gradually into skin-like epithelium laterally with fine hairs on the surface. The posterior limit of the mass could not be defined, as its thick pedicle extended subconjunctivally into the orbit (Fig. 1). Eye movements were free and full. The upper eyelid was incomplete, ending abruptly at the mass and without any

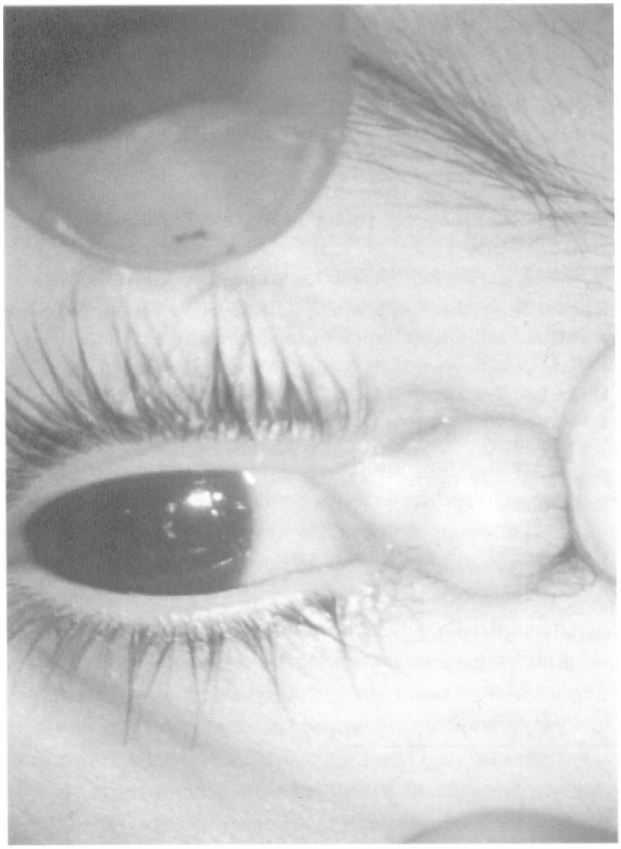

Fig. 1. *Left eye showing the pedunculated tumour occupying the lateral canthus and the upper lid ending on it. Note the fine hairs on the temporal aspect.*

evidence of a lateral canthal ligament or any point of insertion. The supero-temporal fornix was absent. There were no clinical features of any oculofacial syndrome.

Surgical correction started with careful marking of the future lateral canthus and measuring the amount of conjunctiva required for creating a normal deep lateral fornix. A medially based conjunctival flap was raised from the mass consisting of the hairless portion. All the hairy portion and the redundant skin were circumscribed. The mass was dissected under ×2.5 magnification and traced deep to the orbital septum. A subtotal excision was performed. Lateral canthus was reconstructed by raising a periosteal flap from the lateral orbit and suturing the upper tarsus to it with 4-0 Mersilene. The conjunctival flap (see above) was anchored to the depth of the lateral orbit to create a deep lateral fornix. Systemic steroids were used post-operatively to limit the swelling. The cosmetic and functional results were excellent.

Pathology

On gross examination the specimen was a pedunculated mass 6 mm across and yellow on cross-section. Histologically, it was covered by stratified keratinised squamous epithelium with plenty of pilosebaceous units and sweat glands. The tumour was composed of mature fat surrounded by dense connective tissue. These features are those of a dermolipoma. Numerous foci of nerves, smooth muscle, skeletal muscle and a focus of bone surrounded by periosteum-like dense connective tissue were also found (Fig. 2).

Comment

Dermolipomas usually present as soft yellow or pink sessile, subconjunctival masses located anteriorly near the temporal fornix. Posterior extension occurs often, when they may be adherent to several orbital structures. Surgical excision should therefore be conservative.[3]

Dermolipomas have been classified as choristomas, i.e. they are formed by continued growth of tissues not normally indigenous to that area. They consist mainly of mature fat covered by stratified squamous epithelium. Rarely, when they contain other mesenchymal derivatives such as cartilage, bone and smooth muscle, they are called 'complex choristomas'.[2]

Coloboma of the eyelid is rare and its pathogenesis is uncertain. According to the studies of Tessier, it is considered as a form of facial cleft.[4] Others have suggested that it is a disruption anomaly due to the pressure of amniotic bands on the developing face.[5] In coloboma with facial clefts, and even in some of the isolated colobomas, the amniotic band theory explains all the observations.[6] For instance, in these cases it is not unusual to find one or more of the following: a shallow or absent superior fornix, adhesions between the cleft and the globe, alopecia of the eyebrow, a less prominent supraciliary border or disturbance of the anterior hairline.[6]

However, small and isolated lid colobomas are more likely to be the result of a localised failure of adhesion of lid folds *in utero*.[7] In these cases there are no stigmata of facial clefting. Eyelids develop as folds from the frontonasal and maxillary processes at 4–5 weeks of gestation. The folds then fuse along their free margins between the 10th and 12th weeks of gestation, breaking down eventually by the 20th week. Any disturbance (such as by the presence of a growing tumour) in that region could conceivably interfere with the fusion or result in premature breakdown leading to a lid coloboma.

It is difficult to be certain as to when a choristoma develops. It is thought to occur as a result of the inclusion of surface ectoderm and mesoderm during the closure of fetal clefts – an event that should occur early in embryogenesis.[1] In a report of two cases of pedunculated complex choristoma with microphthalmia, the authors suggest that the tumour might have been present at least before the fifth month of gestation and might have interfered with ocular development leading to microphthalmia in one of their cases.[8] In the clinical photograph of the same case a shallow coloboma of the upper lid is seen clearly overlying the pedicle of the tumour. In another report of a pedunculated complex choristoma associated with naevus sebaceus, a coloboma of the upper lid directly over the pedicle is clearly visible in the photograph, although this fact has not been mentioned by the authors.[9] In the same case, the authors

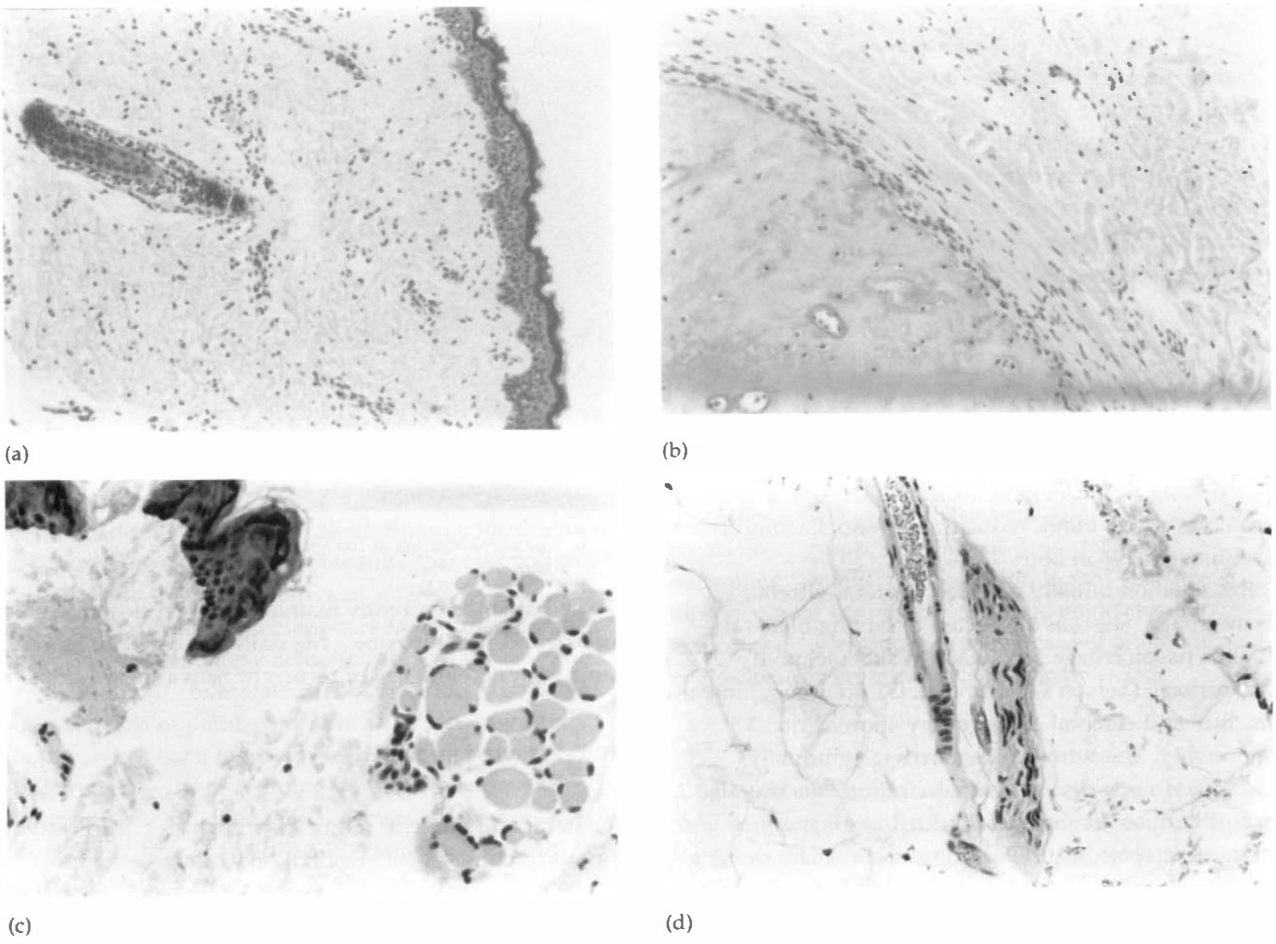

Fig. 2. *(a) Section of the tumour showing keratinised stratified squamous epithelium, dermis-like dense connective tissue and a hair follicle. (b) Central part of the tumour showing a focus of normal bone (lower left) surrounded by periosteum-like dense connective tissue. (c) High-power view showing a bundle of skeletal muscle in cross-section (right). (d) A nerve bundle and a blood vessel in longitudinal section (centre) amidst normal adipocytes. (H&E; original magnifications: ×100 (a, b) and ×200 (c, d).)*

state that 'the base of the mass was adherent to the fornix and inner part of the superior eyelid'. In yet another recently published report, the pedicle of a pedunculated complex choristoma was lodged in a notch in the lateral canthus, thus attesting to the same possibility.[10]

In the present case the child had no features of facial clefting. The lid seemed to end abruptly on the pedunculated tumour. The tumour physically occupied the space of the missing segment of the upper lid and the lateral canthal structures were absent. It therefore seems reasonable to postulate that the developing tumour antedated and interfered with the development of the lid *in utero*, resulting in this rare combination of disorders.

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