BILATERAL ORBITAL CYSTS OCCURRING IN A BANTU INFANT

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and

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Orbital cysts are rare and, clinically, often present diagnostic problems.

The following case with bilateral giant orbital cysts is presented because no similar case can be found in the literature.

CASE REPORT

J.R., African child aged 8 months, admitted to Baragwanath Hospital, 2 September 1952.

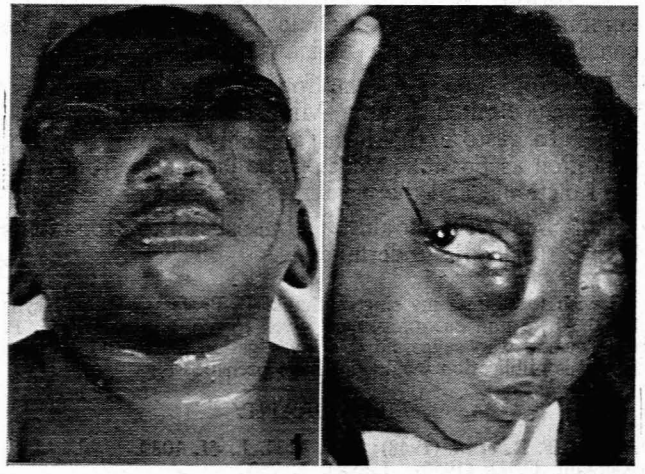
The mother noticed a swelling over the bridge of the nose 6 months ago. At first the swelling was firm, but gradually it spread across the mid-line on both sides of the nasal bridge and became cystic. As the growth progressed it involved both eyes in turn, pushing them upwards and outwards. The child has to move its whole head in order to see an image.

Birth was normal. Since birth the general condition has been satisfactory and progress otherwise normal.

Examination revealed a well-behaved child, with prominent bilateral proptosis. The eyeballs were displaced laterally and there was a marked divergency of the visual axes. There was also a swelling in the midline, which bulged on coughing (see Figs. 1 and 2).

The ophthalmologist's report (I.B.T.) was as follows:

Orbits. Clinically the orbits appear slightly enlarged. There is marked swelling of the orbital tissues which is bulging the lids forwards and has caused the eyeballs to be pushed markedly laterally and forwards.



Figs. 1 *and* 2. Child as it appeared before operation. Note the large cysts medially with the marked displacement of the eyeballs.

Eyeballs. There is an extreme divergent strabismus due to displacement and rotation of the eyeballs laterally. The cornea is clear on both sides. The pupils are equal and react well. The fundi show bulging masses under the retinae on the nasal sides, giving the appearance of detachment of the retina, but due to indentation of the sclera. The discs are slightly distorted and show evidence of stretching of the optic nerves.

Aspiration of Cysts

On 9 September the cyst in the mid-line between the nasal bones was aspirated and cerebrospinal fluid was obtained. This was replaced by 10 c.c. of air, which on X-ray was found to be in the subarachnoid space. This central cyst was diagnosed as a meningocele.

On 15 September the orbital cysts were aspirated. The removal of 28 c.c. of clear yellowish fluid from the right cyst caused no deflation or softening of the left cyst. The left cyst contained 20 c.c. Air was inserted and the patient was X-rayed. The report (16 September) of the South African Institute for Medical Research on the cyst fluid was as follows:

> 'Total protein 0·9 g. per 100 c.c. Microscopic examination of the centrifuged deposit from this specimen showed a moderate number of erythrocytes and a few polymorphonuclears per high-power field.'

After aspiration the eyes were normal on examination but were somewhat sunken in large fat-free orbits. The cysts rapidly filled up and on the next day they were as tense as before the aspiration.

Operations for Removal of Cysts (S.S.)

The left cyst was removed under general anaesthesia on 16 October 1952. Anterior orbitotomy. Left medial canthotomy. The conjunctiva was cut on the nasal side, exposing a tense cystic tumour. The cyst tracked medially to the eyeball, between the muscle cone and the medial orbital wall, and was attached to the optic nerve. It extended as far posteriorly as the optic foramen. No defect of the orbital walls could be detected. The cyst was enucleated. The inferior oblique muscle was cut and re-sutured later. The conjunctiva was sewn and a dental dam drain was inserted. The canthotomy was repaired. The drain was removed on 18 October.

The right cyst was removed on 3 November 1952 by a procedure similar to that on the left side.

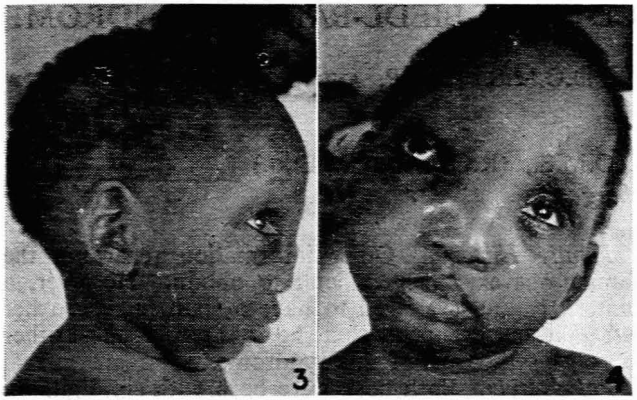
Except for slight orbital oedema the post-operative period was uneventful.

Histology. The cyst-wall showed the structure of oedematous granulations and fibrous tissue. No epithelial lining was observed. The origin of the cyst could not be determined.

Progress. After the operation the eyes resumed their normal straight position, but were somewhat unsupported owing to the lack of orbital fat. Vision appears normal, as the child is able to pick up small objects even with one eye occluded.

Duke-Elder [1] classifies orbital cysts as follows:

1. *Congenital cysts:* (*a*) Dermoids, (*b*) Cephaloceles, (*c*) Congenital cystic eyeball.
2. *Implantation cysts:* following penetrating orbital trauma.
3. *Haematic cysts:* following an intra-orbital haemorrhage.
4. *Parasitic cysts:* e.g. hydatid, cysticercus, filaria.



Figs. 3 *and* 4. Post-operative appearance of the child. The eyes are somewhat unsupported in the large fat-free orbits.

5. *Serous cysts:* hygromata.
6. *Mucous cysts:* from the para-nasal sinuses.
7. *Dental cysts:* invading the orbit from the antrum.
8. *Cysts of intra-orbital organs:* lacrimal gland (dacryops).

The case presented here in a Bantu infant appeared to be one of bilateral haematic cysts, most probably following haemorrhagic disease of the newborn.

The first description of haemorrhagic disease of the newborn, was that of Minot [2] (1852). The disease is characterized by an unusual tendency to spontaneous haemorrhage during the first week of life, usually 2nd-5th day. Townsend [3] in 1894 used the term 'haemorrhagic disease' to describe all cases of haemorrhage in the newborn in which no obvious cause could be found. This excluded cases due to trauma, sepsis, congenital syphilis, and blood dyscrasias such as haemophilia, congenital leukaemia and congenital thrombocytopenia.

Two cases of haemorrhagic disease of the newborn, presenting as unilateral proptosis, were described by Kessel and Williams.[4] In one of their cases there was a low prothrombin index, but the other case had normal haematological findings. The proptosis became less marked after a few days, and in one case had completely disappeared by the end of the third week.

Bilateral exophthalmos in the newborn following spontaneous delivery was reported as a rare occurrence by Kundert.[5] Fracture of the orbit with strangulation of the ophthalmic vein in the superior orbital fissure, retro-bulbar haemorrhage, subperiosteal haemorrhage, and injury of the cavernous sinus, have been considered as aetiological factors in previously reported cases (Edgerton,[6] Windham,[7] Harley [8]). Trauma produced by moulding of the head in its passage through the birth canal was believed to be a primary factor in all cases. The proptosis usually disappears as the haemorrhage absorbs, but may persist or even increase if the haemorrhage becomes encysted.

Pathology of Haematic Cysts. Retro-bulbar haemorrhage usually absorbs slowly owing to the poor blood-supply of the orbital fat. Another factor retarding absorption is the increased pressure in a closed space, which prevents the normal capillary dilatation so essential for hastening absorption. Very rarely the haematoma develops into an encapsulated cyst, remaining as an orbital tumour, which is known as a haematic cyst. The characteristic feature of this kind of orbital cyst is the absence of epithelial and endothelial lining. Structurally they have two coats, an outer fibrous layer, and an inner layer of granulation tissue, rich in capillaries and in which there may be foam cells loaded with lipoids. They contain yellowish or reddish-green fluid and degenerated erythrocytes. The cysts may be situated subperiosteally or in the orbital tissue-spaces.

Similar cysts occur in the breast in localized areas of fat necrosis.

SUMMARY

Bilateral orbital cysts, in themselves, are extremely rare. The condition affecting this child is difficult to classify since it does not fall in with the normal classification of these cysts. The lack of an epithelial lining makes a histological diagnosis difficult. In the absence of a reliable history we feel that the most probable diagnosis is bilateral haemorrhagic cysts; the lack of endothelial lining tends to support this contention. The cystic fluid is most likely a transudate from the granulation tissue, mixed with break-down products of the haemorrhage.

An anterior meningocele, itself a rare finding, was present as well.

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