CASE REPORT

Massive iatrogenic orbital cysts following glaucoma drainage implant and strabismus surgery

Elif Demirkilinc Biler, Ayse Yagci, Onder Uretmen, Melis Palamar

Department of Ophthalmology,
Ege University, Izmir, Turkey

Correspondence to

Dr Melis Palamar,
melispalamar@hotmail.com

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SUMMARY

We describe 2 paediatric patients who developed large orbital conjunctival epithelium-lined inclusion cysts postsurgery. Case 1 underwent Ahmed glaucoma valve implant surgery 8 months ago and case 2 underwent strabismus surgery 7 years ago. Both cases had either symptomatic strabismus or a mass causing some degree of proptosis or incomitance due to lesions in the intraorbital space. Following total excision of the cysts all associated symptoms were resolved. Conjunctival cysts following ocular surgery can develop anywhere in the anterior segment; however, large orbital cysts following anterior segment surgery are quite rare. We think that implantation cysts should be considered in the differential diagnosis of patients that have signs of orbital mass with a history of previous anterior segment surgery. Complete excision of the cysts results in resolution of all associated signs and symptoms.

BACKGROUND

Conjunctival epithelial inclusion cysts are not uncommon and can develop following trauma or ocular surgeries such as strabismus surgery, oculopalpebral surgery, enucleation, and scleral buckling surgery. These cysts have been reported to originate from dislocated epithelium below the surface of the conjunctiva following trauma or during ocular surgery.^{1 2} The cysts are often located in the periorbital region, including the conjunctiva, caruncle and eyelids, and can be detected via slit lamp examination;³ however, large epithelial inclusion cysts extending into the orbit rarely occur. These lesions cause symptoms similar to those associated with orbital tumours. Cases have been reported, primarily as a rare and late complication of strabismus surgery.^{3–7} The incidence of conjunctival cysts following an otherwise uncomplicated strabismus surgery was reported to be 0.25%.⁸ Moreover, formation of orbital conjunctival inclusion cysts following various oculopalpebral surgeries has been reported in adults.^{9–12} To the best of our knowledge orbital inclusion cyst formation following drainage implant (Ahmed glaucoma valve (AGV)) glaucoma surgery and posterior migration of the AGV have not been previously reported.

We report two paediatric patients with intraorbital epithelial inclusion cysts following strabismus surgery (n=1) and AGV implantation surgery (n=1) that caused mechanical restriction of eye movement due to mass effect and proptosis. Ocular history, ophthalmic findings, surgical management, clinical course, and presurgical and postsurgical orbital MRI findings are discussed in detail.

CASE PRESENTATION

Case 1

A 2-year-old girl presented with proptosis and decreased vertical gaze in the right eye. She had a history of surgery for bilateral congenital cataract at the age of 3 months. During follow-up, right aphakic glaucoma had developed and both endoscopic cyclophotocoagulation and glaucoma surgery were performed 2 months apart at another ophthalmology clinic. An AGV implantation was performed in a glaucoma surgery performed 8 months ago elsewhere. Based on anamnesis, no perioperative or postoperative complications were reported.

During primary gaze the right globe exhibited downward dystopia. She had a steady fixation and good ocular pursuit of moving test objects with that eye. She had a low intraocular pressure (IOP) without any medication. Motility examination showed restriction in both upper and, particularly, lower gaze in her right eye. The tube of the AGV was not observed in the anterior chamber and via slit lamp examination no mass was observed in anterior ocular tissues, including the fornices. MRI showed a 19×16 mm extraconal cystic lesion involving the lateral and superior quadrants next to the globe. The cyst had a smooth wall and homogeneously reflective contents, and pushed the globe downwards. At the periphery of the cystic lesion there was a hyporeflexive object considered to be the AGV (figure 1A–C).

Anterior transcutaneous orbitotomy was performed under general anaesthesia. Stallard-Wright incision was used to gain access to the subperiosteal space and the extraconally located cyst. The cyst's contents were stained with methylene blue and the walls were dissected from orbital tissues using blunt manoeuvres. Total excision of the cyst was performed and the AGV implant, which was in the cyst but not in contact with the globe, was also explanted (figure 2). Postsurgery proptosis resolved and there was no longer any gaze restriction or vertical deviation. Histopathological examination of the cyst showed that it was an epithelial cyst. IOP in the right eye was still normal with no medication and no cystic lesion was observed via postoperative follow-up MRI (figure 1C, D), and at 1 year postsurgery the eye remained cyst-free.

Case 2

An 11-year-old boy presented with proptosis, a mass lesion in the nasal conjunctiva of the left eye that was increasing in size, and a decrease in left eye motility over a period of 1 year. He had

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Figure 1 Preoperative and postoperative MRIs of case 1. (A,B,C) Right orbital mass is apparent in the superolateral side of the globe displacing it downwards into the coronal and sagittal sections in preoperative images. A hyporeflective object considered to be the Ahmed glaucoma valve adjacent to the lesion. (B and C) No residual lesion is seen in postoperative control MRI (D and E).

esotropia of +30 prism diopter (PD), hypotropia of 16 PD, and decreased adduction and upgaze in the left eye (figure 3A). Diplopia was not present, probably due to the slow growth of the lesion and development of suppression. There was prominent proptosis in the left eye based on Hertel exophthalmometry (right eye: 12 mm; left eye: 17 mm) and a cystic mass was observed in the left nasal area via gross and slit lamp examinations. Best-corrected visual acuity in both eyes was 1.0 via Snellen chart. Anamnesis was remarkable for esotropia and uneventful strabismus surgery (bimedial rectus recession) at age 4 years. Orbital MRI showed a well-defined $3.5 \times 1.4 \times 1.8$ cm

Figure 2 Peroperative images of case 1. Anterior transcutaneous orbitotomy was performed to reach the subperiosteal space and the extraconally located cyst. Total excision of the cyst was performed. The AVG implant was observed above the cyst but not in contact with the globe (black arrow).

intraorbital cystic lesion that extended into the superonasal quadrant. The cyst was integrated with the left medial rectus, involved the area of insertion, and covered the globe superomedially, which caused temporal and downward displacement of the globe (figure 4A–C). Subconjunctival and intraorbital inclusion epithelial cyst was considered the most likely diagnosis, and excisional biopsy was performed under general anaesthesia. Following conjunctival incision, the cyst wall was bluntly dissected from the surrounding tissues towards the orbita, as much as possible and the cyst's contents were stained with methylene blue to make it more apparent. During surgery the superior portion of the medial rectus was observed to be partially attached to the cyst; as such, isolation of the medial rectus muscle was performed, no suture material was observed and following dissection the cyst was excised totally without any harm to the extraocular muscles (figure 5).

Postsurgery proptosis was completely resolved (13 mm based on Hertel exophthalmometry) and ocular movements had improved (figure 3B). Histopathological examination showed that the cyst was an epithelial cyst. Follow-up MRI in early postoperative period showed no residual cystic lesion (figure 4D, E), and at 1 year postsurgery his findings remained stable. A reoperation for residual esotropia has been planned.

DISCUSSION

Epithelial inclusion cysts are filled with clear serous fluid that contains shed cells or gelatinous mucous materials. The cyst wall in most cases includes several layers of non-keratinising epithelium and connective tissue.¹³ Epithelial inclusion cysts are categorised as primary or secondary/acquired, according to their aetiology. The secondary/acquired type usually occurs as a result of detachment of a part of conjunctival epithelium due to

Figure 3 Preoperative (A) and postoperative (B) photographs of case 2, showing ocular movements in nine diagnostic positions of gaze. The cystic mass was apparent in the superonasal portion of the left globe beneath the conjunctiva in the preoperative photographs.

Figure 4 Preoperative and postoperative MRIs of case 2. (A and B) Left intraorbital mass causing temporal and downward displacement of the globe, observed in coronal sections. (C) The lesion was extended into the superonasal quadrant, covering the globe superomedially, seen in the transverse section. (D and E) No residual lesion is seen in post-operative control MRI.

surgery or trauma.¹⁴ These acquired cysts are benign and can be observed even years after surgery.^{4 5 15}

We presented two paediatric patients with large, intraorbital conjunctival epithelium-lined inclusion cysts, and their MRI findings and clinical management. Both patients developed the

cysts as a complication of ophthalmic surgical procedures that included conjunctival incision. We think the cysts in the presented cases most likely developed as a result of epithelial remnants captured by sutures or the suture needle. It was reported that such intraorbital cysts can also develop due to viable

Figure 5 Peroperative images of case 2. (A) Conjunctival incision, (B) the cyst wall was bluntly dissected from the surrounding tissues towards the orbita. (C) Methylene blue was injected inside the cyst to stain it. (D) The superior portion of the medial rectus was observed to be partially attached to the cyst.

epithelial cells being dragged into the subdermal, subconjunctival, or episcleral space, and the associated inflammation.^{3–5} In some cases the cystic mass may be undetectable, even when carefully performing slit lamp examination.¹⁵ In suspected cases, orbital imaging is useful for diagnosing cysts located in the orbital space, differentiating cystic lesions from solid tumours, and planning and performing surgical excision.^{5 16} Management varies from local excision to observation, based on symptoms, localisation, and type of cyst.¹⁶ There are also thermal cautery and marsupialisation methods described in the literature in the management of these cysts particularly the smaller ones.^{17 18} A mass that causes disfigurement, strabismus, restriction of ocular movement, pressure exerted on the globe, proptosis, or any combination of these (as in the presented patients) is an indication for surgery. Complete excision of such cysts is curative.^{4–6 15 19 20} In cases with involvement of the rectus muscles special care and extensive dissection are necessary, so as not to cause any muscle damage.¹⁵

In addition to following strabismus surgery, conjunctival epithelial inclusion cyst formation can also occur following glaucoma surgery; however, intraorbital cyst formation is rare in paediatric patients. Although AGV implantation is reported to be relatively safe,²¹ case 1 is unusual, as the AGV migrated into the cyst towards the orbita. To the best of our knowledge case 1 is the first patient to be reported with intraorbital epithelial inclusion cyst formation combined with displacement of an AGV following glaucoma surgery.

In conclusion, although rare, intraorbital implantation cysts can cause significant complications and should be considered among space-occupying lesions in orbita, particularly in patients with a history of ocular surgery. In cases in which the posterior border of the cyst cannot be observed via anterior segment examination, orbital imaging can facilitate detailed assessment of the borders and surgical planning. Based on the two cases presented here, we think all ophthalmologists must be made aware of postoperative intraorbital cyst formation in paediatric patients.

Learning points

- ▶ Intraorbital implantation cysts can cause significant complications and should be considered among space-occupying lesions in orbita, particularly in patients with a history of ocular surgery.
- ▶ In cases in which the posterior border of the cyst cannot be observed via anterior segment examination, orbital imaging can facilitate detailed assessment of the borders and surgical planning.
- ▶ All ophthalmologists must be made aware of postoperative intraorbital cyst formation in paediatric patients.

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Contributors EDB contributed to the data collection, surgery, and writing of the manuscript. AY contributed to the data supervision, surgery, and writing of the manuscript. OU contributed to the writing of the manuscript, and surgery. MP contributed to the study supervision and writing of the manuscript.

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