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The formation of epithelial orbital cysts secondary to orbital implants and reconstruction with computer-assisted manufactured patient-specific implant: A two-part case series

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SUMMARY

Epithelial orbital cyst formation is an uncommon sequela to the repair of orbital floor fractures with alloplastic implant materials. This series depicts two cases of orbital cysts associated with a history of traumatic orbital floor fracture and reconstructive surgery. We demonstrate techniques for managing post-traumatic orbital cysts by use of 3D-planned patient-specific implants (PSIs) and surgical navigation (SN).

Keywords: alloplastic implant, orbital fracture, orbital cyst, patient specific implant, surgical navigation.

INTRODUCTION

Alloplastic implants have been widely used in the management of orbital floor fractures. The fracture pattern depends on the mechanism of injury and plays a part in dictating the materials and surgical techniques used to repair the defect. Examples of implant materials include porous polyethene, silicone and titanium (1). However, these can undergo infection, migration, exposure, or more rarely, lead to the growth of orbital epithelial cysts, resulting in significant functional and aesthetic deficits. Treatment of epithelial orbital cysts typically involves cyst excision and implant removal (2). Here, we present two cases of post-traumatic orbital cysts secondary to reconstructive implants, managed jointly by Oral and Maxillofacial Surgery (OMFS) and Ophthalmology using computer-assisted surgical techniques.

CASE 1

A 62-year-old female with a previous right orbital floor fracture presented to the joint Orbital-OMFS clinic 20 years post-injury with right globe dystopia. She sustained the fracture in 2000 following multiple punches to the face and reported diplopia on forward gaze. The fracture was repaired at the time by open reduction internal fixation (ORIF) with placement of a periorbital silastic sheet implant. On postoperative review, she had an interpupillary height difference of 2 mm and persistent diplopia in all fields except on forward gaze. Her diplopia was resolved two months later. In 2015, she was referred to the Ophthalmology and Neurology departments regarding a sudden onset of right hypoglobus and suspected third cranial nerve palsy. She reported an 'asymmetry of her gaze' since her surgery. A palsy was ruled out and she was discharged shortly after an improved condition. She was later referred to Oculoplastics in 2020 for a right dermatochalasis with an associated reduction in visual fields, for which she underwent a blepharoplasty. A marked displacement of the right eye was noticed intra-operatively, prompting further investigation. Medically, she had a background of hypertension, hypercholesterolaemia and asthma. Clinically, there was a 3 mm inferior scleral show and 3 mm upward and forward displacement of the right globe. There was a full range of eye movements, and no diplopia or paraesthesia.

Investigations

A series of CT orbits showed progressive changes around the implant (Figure 1). There was a high-

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Fig. 1. From left to right: coronal slices of a series of CT orbits taken in 2015, 2020, and 2022, respectively.

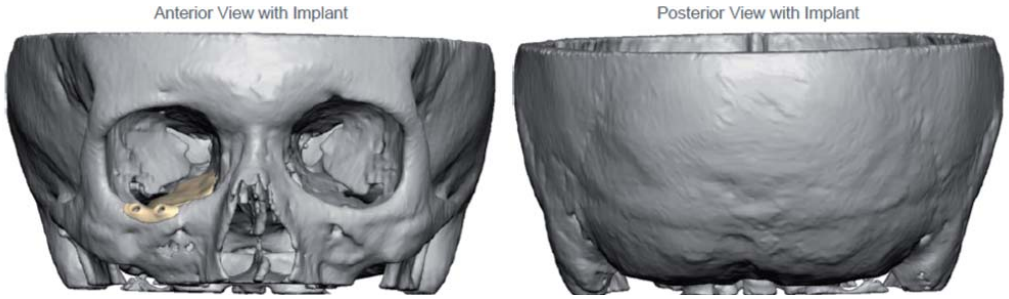

Fig. 2. Surgical plan of the polyetheretherketone patient specific implant in-situ (anterior and posterior views) for reconstruction of the right orbital floor.

Fig. 3. Side-to-side comparison of the patient's right eye pre-operatively (image on the left) and post-operatively (image on the right)

density extraconal cystic collection in contact with the right inferior rectus and globe. Inflammatory changes were present in the right maxillary antrum and there was thickening of the right inferior rectus.

A CT orbit 2015 showed thickening of the right inferior rectus anteriorly and complete opacification of the left maxillary antrum, anterior ethmoid cells and left frontal bone loculus.

A further CT orbit from 2020 showed increased soft tissue or fluid between the repair mesh and the bony floor. The mesh had become superiorly convex, reducing the infraorbital volume. Bilaterally, the intraocular muscles appeared large, with the right lateral inferior rectus being the largest and in direct contact with the mesh. There was persistent opacification on the left, consistent with the previous scan. There were some inflammatory changes in the right maxillary antrum and one of the right anterior ethmoid cells. With these findings put into the context of clinical assessment, evidence suggested a cystic structure in the right eye, which was visible but more minor in the previous scan.

A final pre-operative CT orbit from 2022 showed changed appearances; the meshwork was no longer superiorly convex, and there was a new extraconal high-density cystic collection in contact with the right inferior rectus muscle and right globe.

Treatment and outcome

The patient underwent orbital cyst enucleation and implant removal, using a transconjunctival approach, and replacement with a polyetheretherketone (PEEK) PSI under the joint-care of the OMFS and Ophthalmology teams in 2022. The CT scans were utilised in the three-dimensional planning report for PSI construction (Figure 2). Peri-operatively, a SN system (BRAINLAB®) was used to guide the seating of the implant. Specific anatomical reference points were calibrated to an up-to-date CT scan to confirm the precise implant position.

Histopathological analysis revealed a pseudocystic space with a fibrous wall and lumen containing haemorrhagic material. There was evidence of cholesterol cleft formation, dystrophic calcifications and chronic inflammation in the fibrous wall. No definitive lining was seen.

The patient was followed up at the joint clinic three months post-surgery. Postoperatively, the proptosis had fully resolved, and the upward displacement was reduced by 2 mm (Figure 3). However, the patient reported ongoing diplopia and restriction on upward gaze. A further CT scan revealed soft tissue around the inferior rectus, potentially representing a fibrotic cause for the restriction. She has since been undergoing intraorbital steroid injections with regular review.

CASE 2

A 54-year-old male presented to the joint Orbital-OMFS clinic in 2015 with a 3-year history of a progressive right hyperglobus. He had an alleged assault ten years prior (hit by a metal bar), resulting in

a right-sided zygomaticomaxillary complex (ZMC) fracture with orbital floor involvement. At the time of injury, he complained of right infraorbital numbness and diplopia, which resolved spontaneously the day after. He subsequently underwent an ORIF with placement of an orbital porous polyethene sheet implant (Medpor®) via a transcutaneous approach. He was penicillin allergic but otherwise fit and well. On examination, there was right-sided hyperglobus, diplopia, infraorbital swelling and paraesthesia.

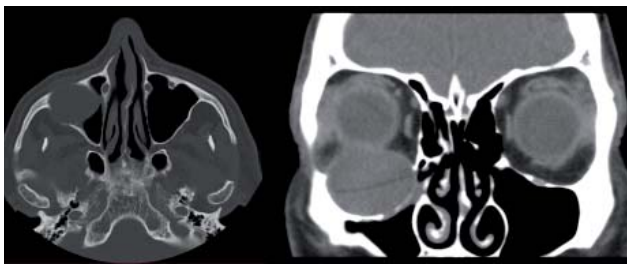

Fig. 4. Transverse and coronal slices of CT orbit taken in 2012

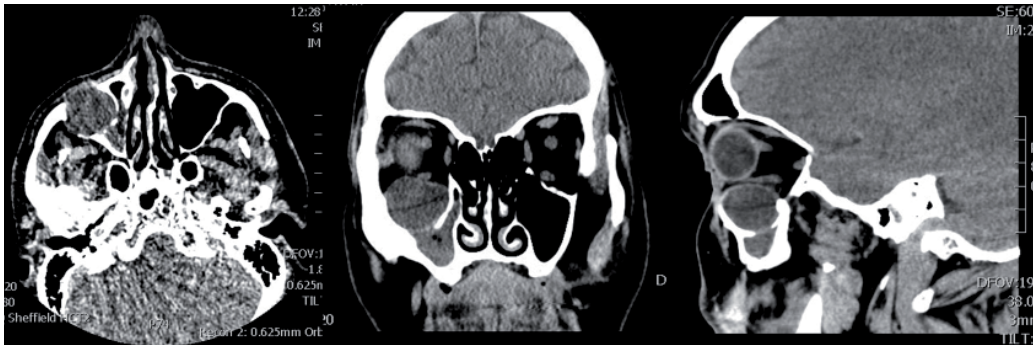

Fig. 5. Transverse, coronal and sagittal slices of a pre-operative CT orbit taken in 2015

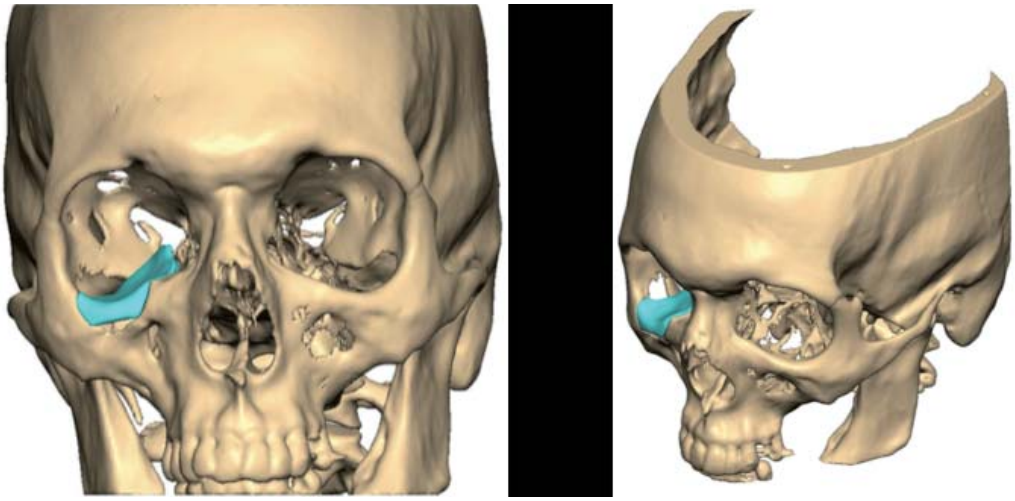

Fig. 6. Surgical plan of the polyetheretherketone patient specific implant in-situ (anterior and lateral views) for reconstruction of the right orbital floor

Investigations

A CT orbit taken in 2012 showed a well-defined, uniform, low-density soft tissue mass pushing the right globe superiorly and anteriorly. There was marked thinning and bowing of the right orbital floor and opacification of the right maxillary antrum (Figure 4).

A pre-operative CT orbit in 2015 showed progressed bowing and thinning of the inferior orbital bony floor by the cyst and soft tissue occupying most of the right maxillary antrum (Figure 5).

Treatment and outcome

The orbital cyst was enucleated, and the previous Medpor® implant removed via a transconjunctival approach. A PEEK PSI was used to reconstruct the orbital floor (Figure 6). Histopathological analysis showed features of a surgical ciliated cyst of the maxilla (Figure 7). It comprised a fibrous cyst wall lined by a ciliated pseudostratified columnar epithelium. In other areas, the lining was attenuated stratified squamous epithelium. The cyst wall contained scattered collections of chronic inflammatory cells.

Post-operatively, the patient reported diplopia at extremes of upward gaze but otherwise had a full range of eye movements and no evidence of enophthalmos (Figure 8). He was discharged following resolution of symptoms.

DISCUSSION

Orbital epithelial cysts may present with pain, paraesthesia, proptosis, hyperglobus, ophthalmoplegia, diplopia and reduced visual acuity (2). It is less well-known how the cysts form. It has been hypothesised that a transconjunctival approach during initial orbital floor repair provides a pathway for epithelial cell migration that goes on to form the cyst lining (3). An environment of chronic inflammation is another potential pre-requisite for cystic growth. A systematic review found no significant association

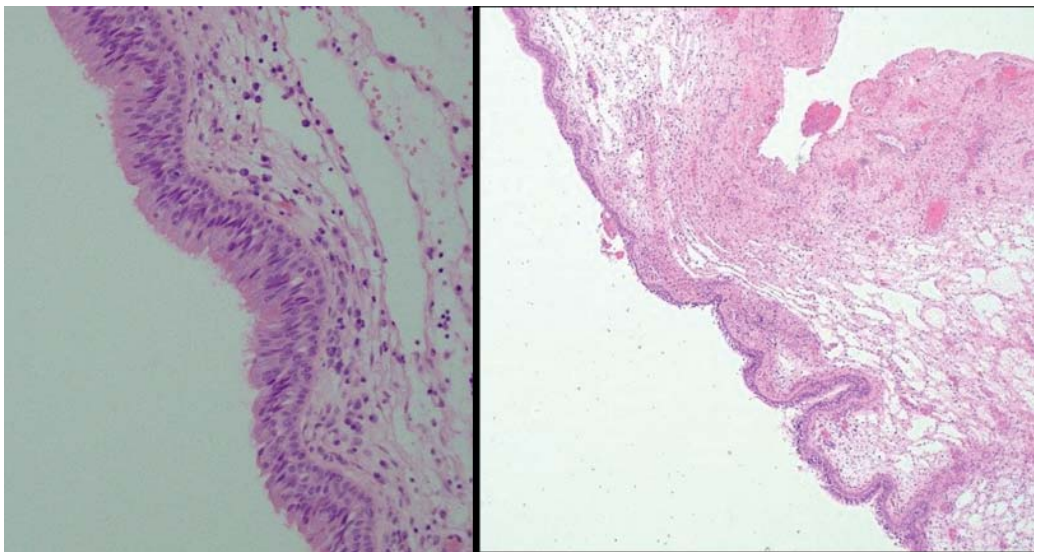

Fig. 7. Histopathological images of the maxillary ciliated cyst (haematoxylin and eosin staining) showing fragments of variably dense and loose fibrovascular connective tissue, partly lined by ciliated pseudo-stratified epithelium. An infiltrate of chronic inflammatory cells comprised of mainly plasma cells is seen within the connective tissue beneath the epithelium and perivascularly.

Fig. 8. Side-to-side comparison of the patient's right eye pre-operatively (image on the left) and post-operatively (image on the right)

between cyst formation and the implant material used. The management of these cysts involved cyst and implant removal with, or without, replacement using an alternative alloplastic implant material or an iliac bone crest graft (2). To prevent globe dystopia following cyst excision we have reconstructed the orbital floors. Complete excision of the cyst wall is considered fundamental in preventing recurrence (4).

We present a case of a pseudocyst detected 20 years after orbital floor reconstruction with a silastic implant and a second case of a true epithelial orbital cyst presenting seven years after placement of a porous polyethene orbital implant. Both were treated with cyst enucleation, implant removal and replacement with PEEK PSIs via the transconjunctival approach. To the best of our knowledge there are no cases of orbital cysts associated with PEEK PSIs and the use of PEEK also allows intra-operative adjustment of the implant.

PSIs have classically been used in complex cases, such as those of structurally irregular defects, volume loss (5), craniofacial involvement or prior unsuccessful repair (6). PSIs have been previously compared to conventional implants. Kim et al. used CT scans to numerically compare the orbital defect pre- and post-surgery. The PSI group had significantly reduced bone defect areas compared to the conventional group (7). Kittichokechai et al. compared the orbital tissue volume and exophthalmometry values derived from CT scans pre- and post-surgery. The mean difference of the parameters was significantly reduced post-operatively (8).

However, PSIs, equally, can be subject to complications. One study had a faulty implant due to the incorrect translation of areas of thin bone to the computer-aided design. Inaccurate implant positioning was another issue (9). We utilised SN, whereby specific anatomical reference points were calibrated to a CT scan at the start of the procedure, and used to confirm the precise implant position in real-time. Averages of <1mm in technical system accuracy and 1-2mm in intraoperative precision have been reported when SN has been used for various surgical applications (10). SN also allows anatomy within the orbital cavity to be defined.

Our cases demonstrate the importance of accurate diagnosis, appropriate surgical intervention, and a multidisciplinary approach to effectively manage post-traumatic orbital cysts. Using adjuncts such as SN and 3D-planning in implant construction may encourage more precise surgical outcomes. Regular follow-up is crucial for the careful monitoring of postoperative outcomes. There may be a place for tailored design and computer-assisted techniques to exist in broader practice. However, further research would be beneficial to refine its use in managing this rare complication.

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