Available online at www.sciencedirect.com

SciVerse ScienceDirect

British Journal of Oral and Maxillofacial Surgery 54 (2016) 658–663

BRITISH Journal of Oral and Maxillofacial Surgery
www.bjoms.com

Epithelial cysts associated with alloplastic implants after repair of orbital fractures: a systematic review and four new cases

Yun Su, Jing Sun *, Xianqun Fan *

Department of Ophthalmology, Shanghai Ninth People's Hospital, Shanghai Jiao Tong University, School of Medicine

Accepted 29 March 2016
Available online 17 April 2016

Abstract

An epithelial cyst is a rare and often late complication of long-term alloplastic implants, which has the potential to lead to further complications and harm to patients. We made a systematic review of papers published during the past 30 years about the mechanisms and clinical characteristics formation of epithelial cysts after repair of an orbital fracture by searching PubMed, Medline, and Web of Science to collect all related case reports and series published in the English language. We also made a retrospective review of casenotes of all patients diagnosed with orbital epithelial cysts in our department. We found 19 cases of epithelial cysts, including the four cases of our own, associated with alloplastic material, 12 of which were associated with silicone. There were 12 men and seven women aged from 26-71 years old. Orbital cysts developed 15 months-31 (median 8) years after implantation. Histological analysis confirmed that the cysts were all epithelial cysts lined with squamous or respiratory (or both) cells, and differing degrees of chronic inflammation. Epithelial cysts after implantation of alloplastic material may present with various symptoms several years after repair of orbital fractures, and their formation probably results from the synergistic effects of both ectopic cells and chronic inflammation. The implant itself may be a trigger, and the cysts did not seem to be limited to one specific type of implant.

Keywords: Epithelial cyst; Orbital fracture; Alloplastic implant; Complication

Introduction

Alloplastic implants, such as porous polyethylene, hydroxyapatite, and silicone, are commonly used for orbital reconstruction to cover and reinforce the bony defect, release the entrapped soft tissue, and restore the natural anatomy and orbital volume.[1] Formation of an orbital cyst is a rare and late complication of alloplastic implants after orbital fracture repair. The cyst may lead to displacement of the implant, and present with proptosis, diplopia, orbital discomfort, or even compression of the optic nerve. A second operation, which is relatively complicated, is usually needed to remove the implant, with more potential complications and increased harm to patients.

Most of the orbital cysts reported are haemorrhagic or haematic cysts,[2,3] and we know of only a few case reports of epithelial cysts associated with alloplastic implants in the orbit.[3,4] We present the results of a systematic review over the past 30 years on the mechanism and clinical characteristics of formation of epithelial cysts after repair of orbital fractures. We have also included four new cases from our department.

* Corresponding authors at: Department of Ophthalmology, Shanghai Ninth People's Hospital, Shanghai Jiao Tong University, School of Medicine, No. 639 Zhizaoju Road, Shanghai, 200011, China. Tel.: +8621-63135606; fax: +8621-63134218.

E-mail addresses: suyun_sy@126.com (Y. Su), sophiasj@126.com (J. Sun), fanxq@sh163.net (X. Fan).

http://dx.doi.org/10.1016/j.bjoms.2016.03.028

Table 1
Patients who presented to us with orbital epithelial cysts.

Case No.	Age (years)	Sex	Site of fracture	Implant used	Interval (years)[‡]	Clinical presentation	Computed tomographic scan	Intraoperative findings	Histological examination
1	55	M	Medial wall	Hydroxyapatite	8	Inferior scleral show, diplopia, exophthalmos, restricted ocular mobility	An inferior 27x20 mm cystic mass in the left orbit. The implant was totally inside the cyst (Fig. 1).	An orbital cyst with clear serous fluid.	Squamous epithelium with chronic inflammation.
2	26	F	Medial wall	Hydroxyapatite	2.5	Restricted ocular mobility, hyperglobus, exophthalmos	A cystic semispherical lesion in the inferomedial orbit above the implant.	An orbital cyst with clear serous fluid.	Squamous epithelium with chronic inflammation (Fig. 2).
3	32	F	Floor and medial wall	Titanium	0.9	Hyperglobus, exophthalmos, pain	A 20x15 mm, spherical and well-circumscribed mass in the left orbit, encapsulating the titanium implant.	An orbital cyst with sanguinopurulent fluid	Stratified squamous epithelium (Fig. 3).
4	38	F	Floor	Medpor®	4	Proptosis, diplopia, exophthalmos, restricted ocular mobility	A 21x32 mm orbital cyst, which was divided into several segments by three visible implants (Fig. 4).	A large orbital cyst with Medpor® implants wrapped inside.	Squamous epithelium

[‡] Interval between implantation of materials and presentation of orbital cyst.

Method

We searched electronic databases including PubMed, Medline, and Web of Science to collect all related case reports and series in the English language before July 2015. We used combinations of the following terms: epithelial cyst, conjunctival or squamous epithelial cyst, respiratory or columnar epithelial cyst, and repair or reconstruction of an orbital fracture. We then made a systematic review. All cases were reviewed and available data collected on age, sex, site of fracture, implant used, interval between implantation of material and presentation of the orbital cyst, clinical presentation, treatment, contents of the cyst, and histological examination of the cyst's wall.

We also retrospectively reviewed the case notes of patients who presented to our hospital with orbital epithelial cysts. Relevant demographic data, clinical presentations, radiological and histological findings, procedures, and follow-up information were collected. This study followed the tenets of the Declaration of Helsinki and was accepted by our Ethics Committee. Informed consent was obtained from patients included.

Results

Four patients were diagnosed with orbital epithelial cysts after repair of orbital fractures in our department (Table 1). In addition, several cases of epithelial cysts in relation to alloplastic materials have been reported elsewhere. Fifteen of these patients, 11 men and four women aged from 28-71 years old (median 49) are described in Table 2.[1–14] The orbital floor or zygomatic complex were the most common sites of fracture, with one case of a Le Fort II fracture, which also involved the orbital floor. Only one case involved the orbital roof.

The clinical presentations included diplopia, proptosis, hyperglobus, and restricted ocular mobility, which were similar to other orbital tumours. Visual acuity was reduced in only one case, in which Hillier et al.[9] found a large inferior orbital mass compressing the globe and the optic nerve.

In 12 cases the implant was made of silicone, in one case porous polyethylene (Medpor®), in one Medpor®/titanium, and in one polytetrafluoroethylene (Teflon®). Orbital cysts developed a median of 8 years (range15 months-31 years) after implantation.