

Case Report

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# Conjunctival Epithelial Inclusion Cyst following Evisceration with Primary Orbital Implantation

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## Keywords

Conjunctival epithelial inclusion cyst · Evisceration · Orbital implant · Excision

## Abstract

A 29-years-old Turkish man who had undergone evisceration with primary orbital implantation 20 months prior complained of difficulty wearing his artificial eye. Slit-lamp examination revealed a conjunctival cyst in the center of the anophthalmic socket, with no evidence of scleral or orbital implant exposure. The cyst was completely excised under general anesthesia and did not require use of any sclerosing substance or dye. At 6 months postoperatively, there was no recurrence of the cyst or exposure of the sclera or orbital implant. As the upper and lower fornices were sufficiently deep, the patient could wear his artificial eye.

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## Introduction

Development of conjunctival cysts is a rare complication in anophthalmic sockets. There are 3 proposed mechanisms for their development: implantation of a free fragment of conjunctiva into deeper orbital space; incarceration of an edge of the conjunctiva during wound closure; and conjunctival dehiscence followed by epithelial downgrowth [1]. Conjunctival cysts occur in 0–7% of patients in whom an orbital implant is inserted [2–6] and more frequently develop after a second implantation procedure [3]. Management for this entity is challenging. Although complete excision and marsupialization of the cysts are the gold standards, these procedures are difficult to perform in most cases. Several treatment options exist in these scenarios. Here, we report a case of complete excision of a conjunctival epithelial inclusion cyst occurring after evisceration with primary orbital implantation.

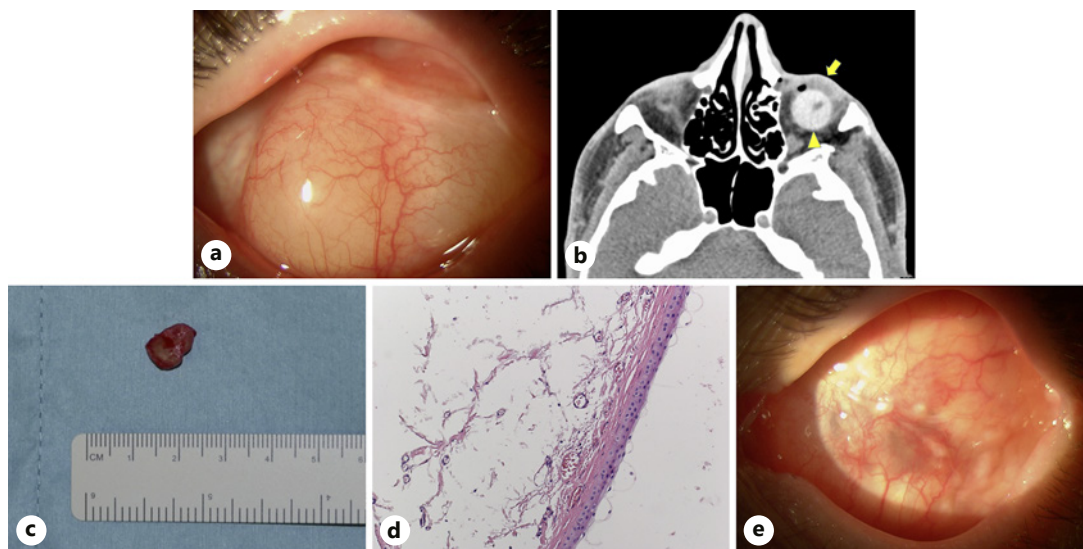
## Case Report

A 29-years-old Turkish man was brought to the emergency room of our hospital suffering from multiple facial bone fractures, cerebrospinal fluid leakage, and left globe rupture following a traffic accident. Two days after the traffic accident, evisceration of the ruptured globe was performed by one of the authors (Y.T.) to lessen the risk of sympathetic ophthalmia [7]. During surgery, 4 scleral flaps were made [8]; a 20-mm acrylic ball was placed into the scleral shell; and Tenon's capsule and the conjunctiva were closed tightly. Postoperative course was uneventful, and the patient was transferred to another hospital at 27 days postoperatively.

Twenty months after surgery, he presented again at our institution due to difficulty wearing his artificial eye. Slit-lamp examination revealed a conjunctival cyst in the center of the anophthalmic socket (Fig. 1a). There were no signs of sclera or orbital implant exposure at this time. Computed tomographic images demonstrated a cyst just anterior to the orbital implant (Fig. 1b). The mass was predominantly hypodense but showed focal hyperdensities. The cyst was completely excised under general anesthesia by one of the authors (Y.T.) (Fig. 1c). Injection of a sclerosing agent or dye was not needed during surgery. No fistulous tracts were found. A small area of sclera was exposed following cyst excision. However, the conjunctiva was easily mobilized to cover this defect. Pathological examination showed stratified squamous epithelium with no goblet cells lining the cyst walls (Fig. 1d). Mucinous materials were also found within the cyst cavity. At 6 months postoperatively, there were no signs of recurrence nor of scleral or orbital implant exposure (Fig. 1e). As the upper and lower fornices were sufficiently deep, the patient could wear his artificial eye.

## Discussion

We report a rare case of a conjunctival cyst that developed following evisceration with primary orbital implantation. Conjunctival cysts are rarely associated with primary orbital implantation. They occur more frequently after secondary implantations because of more complicated



**Fig. 1.** Case presentation. **a** Slit-lamp examination 20 months after evisceration showing a cyst at the center of the socket. **b** The cyst (arrow) is located anterior to an orbital implant (arrowhead) on an axial CT image. **c** The cyst was completely excised. **d** The cyst is lined by stratified squamous epithelium without goblet cells (hematoxylin and eosin staining; magnification,  $\times 200$ ). **e** No recurrence of the cyst was found at 6 months postoperatively. CT, computed tomographic.

procedures and tight Tenon's capsule, and the conjunctival layers increase the chances for conjunctival migration into a deeper orbital space [3]. While the causes for conjunctival migration in this case were not clear, there were no signs of conjunctival dehiscence or a fistulous tract. It is thus highly likely that a small fragment of conjunctivae may have been implanted during evisceration, resulting in the development of a conjunctival epithelium inclusion cyst [1].

While complete excision was achieved in the present case, this is often difficult because cyst walls are delicate and easily rupture at any point during surgery [9, 10]. Identification of ruptured cyst walls is difficult [9, 10]. On the other hand, complete excision can also lead to socket contracture and deterioration of socket movement and cosmetic outcome [11, 12]. Injection of a fibrin sealant into an intact cyst has been suggested to aid in en bloc excision, but this method is associated with an increased risk for viral transmission [9]. Injection into intact cysts with absolute alcohol [10], trichloroacetic acid [12–14], or sodium tetradecyl sulfate [11] has also been proposed. In the case of ruptured cysts, a study proposes using indocyanine green to stain inner cyst walls and aid in identification [15].

The remaining conjunctivae were adequate to sufficiently cover the wound, and no patches were necessary in this case. However, in cases where there are inadequate conjunctivae, a skin flap can be created from the upper eyelid or lateral canthal area and mobilized over the defect.

Enucleation had been conventionally performed in eyes with penetrating ocular injury to minimize the risk of sympathetic ophthalmia. However, a recent study showed that evisceration reduces the risk of sympathetic ophthalmia to a similar degree [16]. In addition, evisceration is associated with shorter surgical times and better postoperative cosmesis [16]. We, therefore, chose evisceration in the present case.

Computed tomographic images showed that the cyst was filled with areas of hyperdensity. This may be due to secreted materials within the cyst cavity.

In conclusion, while conjunctival cysts are a rare complication of evisceration with primary orbital implantation, ophthalmologists should be aware of this entity, as well as be knowledgeable of the treatment options available for proper management.

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## Statement of Ethics

The authors adhered to the tenets of the 1964 Declaration of Helsinki. Written informed consent was obtained from the patient and patient's spouse for publication of this case report and any accompanying images. We asked the institutional review board of Aichi Medical University Hospital and confirmed that the ethics approval for this report was not necessary on the basis of the ethical guidelines for medical and health research involving human subjects established by the Japanese Ministry of Education, Culture, Sports, Science, and Technology and the Ministry of Health, Labor, and Welfare.

## Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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## Author Contributions

All the authors qualify for authorship based on contributions to the conception and design (Y.T.), acquisition of data (Y.T.), literature search (Y.T.), and analyses and interpretation of data (all authors). All the authors contributed to drafting the article and revising it critically for important intellectual content and final approval of the version to be published.

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