

Conjunctival Squamous Cell Carcinoma due to Long-term Placement of Ocular Prosthesis

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Summary: Conjunctival squamous cell carcinoma (SCC) arising from an anophthalmic socket is quite rare, with few reports in the English literature. A 59-year-old man who had used an ocular prosthesis for 40 years had not removed the ocular prosthesis at all during the last 5 years. He had developed a mass on his entire right upper eyelid, and biopsy revealed a moderately differentiated SCC. Orbital exenteration including the upper and lower eyelid skin was performed. The defect was reconstructed with a free forearm flap followed by the placement of a facial epithesis. The pathology revealed an intraepithelial carcinoma on the upper palpebral conjunctiva, which seemed to infiltrate exclusively from that site to the upper eyelid and into the orbit. Other risk factors were not detected; therefore, chronic irritation or microtrauma of the upper conjunctiva from the prosthesis due to persistent prosthesis placement could have been the main trigger for the development of SCC. In cases where the ocular prosthesis is not fitted properly or removed appropriately, clinicians should be aware of this possible long-term consequence. (*Plast Reconstr Surg Glob Open* 2015;3:e325; doi: 10.1097/GOX.0000000000000299; Published online 16 March 2015.)

The incidence of squamous cell carcinoma (SCC) of the conjunctiva is 0.02–3.5 per 100,000 people, depending on the specific geographical location.¹ This condition typically presents in the elderly, white man and is more likely to occur in the limbal or bulbar areas.² The most common risk factor for conjunctival SCC is excessive exposure to ultraviolet radiation; however, spending more than 50% of the time outdoors during the first 6 years of life while living within 30° latitude of the equator is also a

risk factor for ocular surface SCC.^{2,3} Newton et al⁴ reported that the incidence of SCC of the eye declined by 49% for each additional 10° latitude away from the equator. The other risk factors for conjunctival SCC tumors are chronic irritation or microtrauma and infectious agents such as Human papillomavirus (HPV) 16 or 18, human immunodeficiency virus, or trachoma.⁵ In contrast, a conjunctival SCC arising from an anophthalmic socket is quite rare; there have been only 10 cases reported in English literature.^{2,5–10}

We herein report a case of a patient who developed conjunctival SCC due to long-term placement of an ocular prosthesis.

CASE REPORT

A 59-year-old man was observed at our hospital for the evaluation of a right upper eyelid mass. He had suffered an ocular laceration injury to his right eye at the age of 8 and the visual acuity in that eye had decreased to light perception. At 18 years of age, his right eye was completely blinded when he suffered another injury. For the past 40 years, the patient had used an ocular prosthesis; however, he had

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not removed it at all over the last 5 years. For the 6 previous months, he had noticed a mass on his right upper eyelid, which had gradually increased in size. Despite visiting his local ophthalmologist and having the ocular prosthesis removed, the eyelid swelling continued to worsen; therefore, he was referred to our department for further examination.

At our evaluation, we had found that the entire right upper eyelid comprised a large, swollen, dark purple protrusion (Fig. 1). The patient was unable to open his right eye and an uneven subcutaneous mass was palpated. There was no pain or malodor, and no cervical or preauricular lymph node swelling was observed.

Magnetic resonance imaging revealed a solid mass that extended over the entire upper eyelid into the eye socket (Fig. 2). Computed tomography findings indicated no bone destruction and no distant metastases. After a biopsy, the lesion was diagnosed as a moderately differentiated SCC. We planned an extensive resection of the tumor.

Considering the spread of the tumor on radiologic imaging, the resection margins were set below the eyebrow for the upper eyelid, the orbital border for the lower eyelid, and 2 cm from the orbital border for the lateral margin (Fig. 3A). We performed orbital exenteration including the periosteum. Because the perma-



Fig. 1. Initial presentation at our hospital. The entire right upper eyelid comprised a large, swollen, dark purple protrusion, and he could not open his eye. There was no pain and the hard, subcutaneous uneven mass was palpitated.

nent preparation results indicated negative margins, reconstructive surgery was performed using a free forearm flap; this was followed by the ocular prosthesis. To create a deep orbital floor, we partially resected the middle of the flap into a wedge-shaped section (Fig. 3B). The resected tissue specimens had indicated that the tumor was a moderately-to-well differentiated SCC and intraepithelial carcinoma was detected on the palpebral conjunctiva near the upper eyelid margin (Fig. 4B). It had infiltrated exclusively from that site to the subcutaneous tissue of the upper eyelid and into the orbit. The iris and lens shapes from the eyeball had been maintained as the tumor had extended over the remaining eyeball tissue (Fig. 4C). No tumor tissue had stained positive for polyclonal antibodies HPV 6, 11, 16, or 18.

The postoperative course was favorable and radiation was locally administered at 50 Gy. Six months postoperatively, an epthesis for the eyelid defect was manufactured for the patient and he returned to his regular job. Five years postoperatively, there has been no recurrence or metastasis and the patient has remained disease free (Fig. 3C).

DISCUSSION

Few reports of conjunctival SCC that has developed in an anophthalmic socket exist within the literature. Here, we have described a case that developed after 5 years of long-term insertion of ocular prosthesis.

Campanella et al⁶ first reported 2 cases of this tumor in 1998. Each of the previously reported cases in the literature had a history of wearing prosthesis for more than 40 years,^{2,5,7-10} except for 1 case that developed an in situ SCC after 32 years of wearing the original prosthesis without routine maintenance.¹¹

Although excessive sun exposure could be a risk factor for developing conjunctival SCC in general, none of them suspected it. Other risk factors such as local HPV infection or systemic immunodeficiency disease did not contribute to these cases; therefore, chronic irritation was speculated as the main contributing factor to the tumor development in all cases. Some of the patients had complained about an ill-fitting prosthesis for a long time.

In our case, instead of having a poor fitting prosthesis, the patient did not remove it for 5 years. Kim et al¹² reported that although the conjunctiva in the anophthalmic sockets showed squamous metaplasia, there was no correlation between the particular aspects of prosthesis care, including total wearing time and frequency of polishing, and cytologic features of the conjunctival epithelium.

Our case had developed intraepithelial carcinoma on the upper palpebral conjunctiva, and the shape of the remaining phthisis bulbi had been maintained without growth of the tumor. Therefore, the tumor seemed

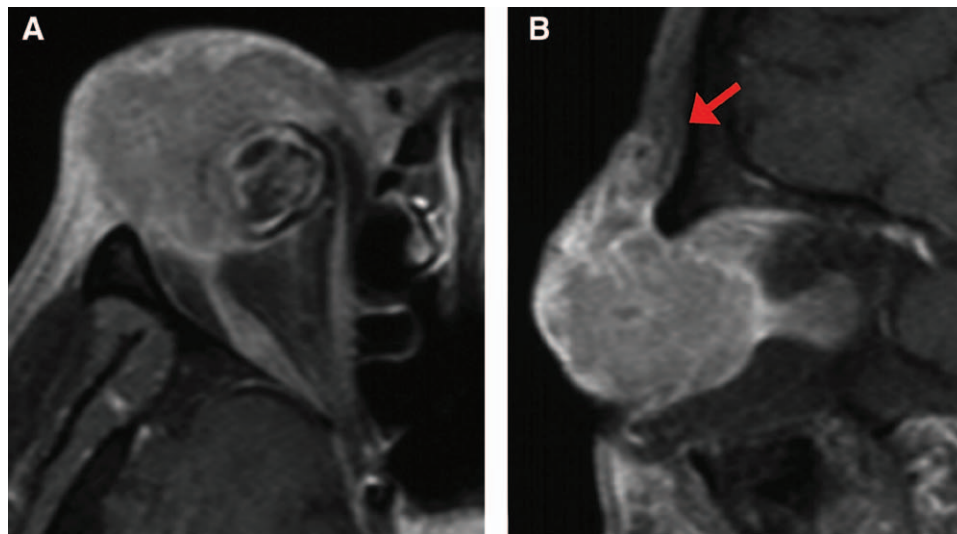


Fig. 2. Magnetic resonance imaging (MRI) images (T1-enhanced; A, axial; B, sagittal). MRI revealing a solid mass with mottled contrast effects extending over the entire upper eyelid into the eye socket. The borders were unclear and the contrast effects followed the skin of the upper eyelid (red arrow). The shape of the mass suggested that it extended from the subcutaneous tissue into the eye socket.

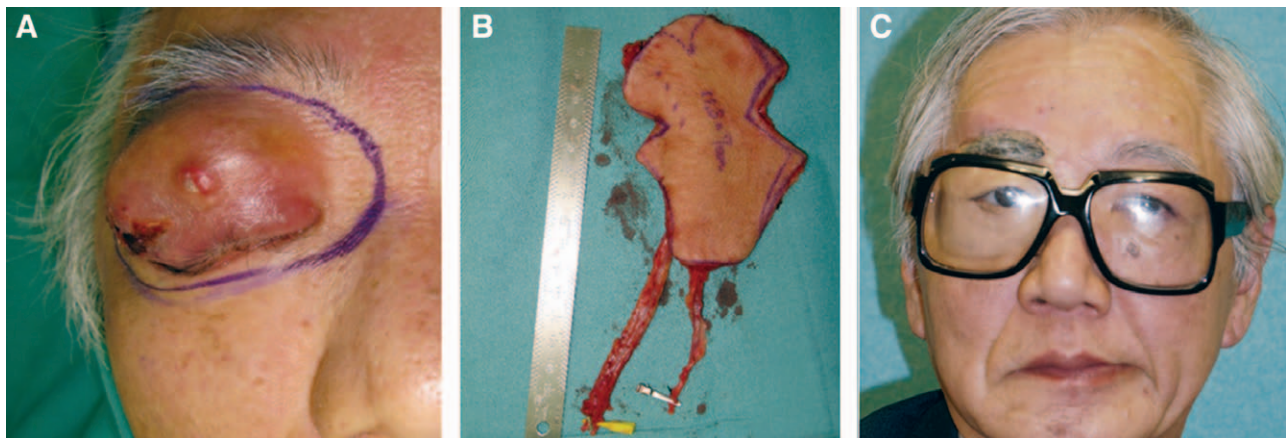


Fig. 3. Intraoperative and postoperative views. A, The design of the tumor excision. The resection margins were set below the eyebrow for the upper eyelid, at the orbital border for the lower eyelid, and 2 cm from the orbital border for the lateral margin. B, Harvested free forearm flap. We partially resected the middle section of the flap into a wedge shape to make it fit into the orbital floor. The flap was transferred by anastomosing the pedicle to the superficial temporal artery and vein. C, Five years after the operation. He has been wearing the epithesis for more than 5 years without any problem.

to infiltrate exclusively from the upper conjunctiva to the subcutaneous tissue of the upper eyelid and into the orbit. As in most of the previous cases, the tumor did not arise from the deep surface of the anophthalmic sockets; our patient did not spend much time under Ultraviolet-B light nor did he have confirmed HPV infection in the excised tissue. Presuming prosthesis care has not been related to carcinogenesis, we also hypothesized that chronic irritation or microtrauma between the upper conjunctiva and the prosthesis could be the main trigger for developing SCC because continuous friction of the prosthesis could easily create these conditions through the daily movement of the upper eyelid.

With respect to the treatment for conjunctival SCC, except for the in situ cases from Barrett et al,¹¹ all cases were treated surgically with or without subsequent radiation. Even in healthy-appearing anophthalmic sockets, the conjunctiva showed squamous metaplasia and had a decreased goblet cell density with an increased nucleus-to-cytoplasm ratio compared with the controls.¹² Therefore, we thought orbital exenteration including the upper and lower conjunctiva would be essential; the upper eyelid skin, which we thought was involved, required total resection.

For the reconstruction of the defect, we chose to use a facial epithesis because both the upper and lower

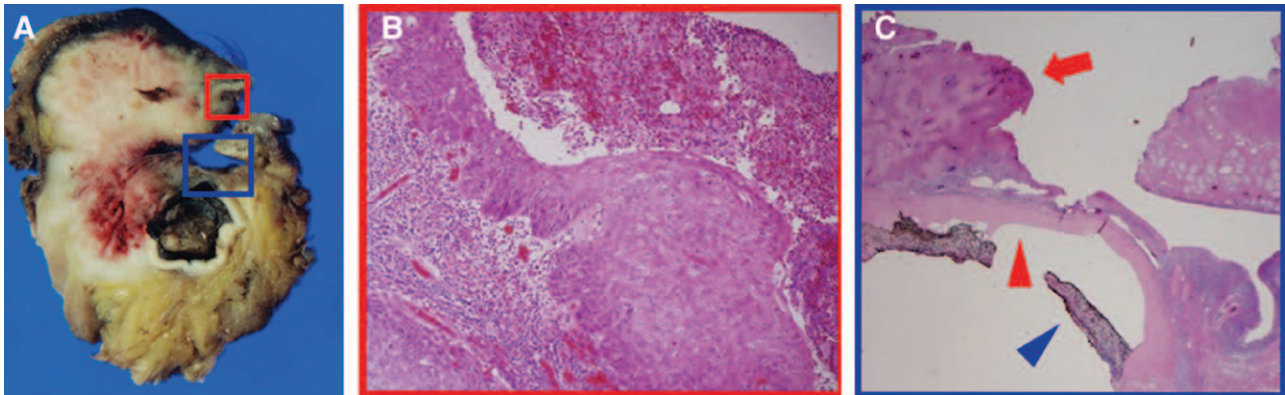


Fig. 4. Pathological findings. A, The macro image of the wide resected specimen (red small square, center area; blue large square, right area). B, Upper eyelid conjunctiva revealed an intraepithelial carcinoma on the palpebral conjunctiva near the upper eyelid margin ($\times 100$). C, In the remaining eyeball area, the tumor (red arrow) extended over the ocular tissue; the iris and lens shapes were maintained ($\times 20$; red triangle, the lens; blue triangle, the iris).

eyelid skin were included in the resection. We planned subsequent radiation therapy, and the patient wanted to recover and return to his job as soon as possible. To use the epithesis firmly, we had to create a deep orbital space. Therefore, partially resecting the middle of the forearm flap into a wedge shape, we could fit the flap into the deep orbital space easily. The patient has worn the epithesis for more than 5 years without any trouble, and he has been free of disease.

CONCLUSIONS

We have described a case who developed conjunctival SCC due to persistent long-term placement of an ocular prosthesis. Conjunctival SCC that arises from an anophthalmic socket is quite rare; however, unlike regular conjunctival SCC, chronic irritation or microtrauma between upper conjunctiva and prosthesis has been suspected to be the main trigger for the development of this disease.

Clinicians should be aware of this possible long-term tumor outcome, especially in cases where the prosthesis has not been maintained adequately.

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PATIENT CONSENT

Patient provided written consent for the use of his images.

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