



Case report repairing orbital skin defects using composite flaps after giant eyelid-derived tumor excision and orbital exenteration

Bin Fan, PhDa, Jian-Ju Liu, PhDb, Bei-Fen Wang, MDa, Ying-Jian Sun, PhDa, Guang-Yu Li, PhDa,

Abstract

Rationale: Though giant malignant tumors arising in the eyelid are rare, they often require extensive surgery for removal along with orbital exenteration. Because of this, repairing orbital defects is an important factor in the surgical strategy.

Patient concerns: Case 1 was a 78-year-old nomad man who presented in the Department of Ophthalmology with a giant tumor in his right eyelid, which had developed over three years. Clinical examination revealed a huge pigmented, nonhealing ulcerated lesion, approximately $52 \times 44 \times 40 \, \text{mm}^3$ in size. Case 2 was a 52-year old rural male complaining of a huge tumor in the right eyelid. Patient medical history revealed that the mass was initially the size of a soybean and gradually grew over 3 years to the size of a fist.

Diagnoses: Histopathological examination of the tumors revealed that one was a basal cell carcinoma and the other a sebaceous gland carcinoma.

Interventions: The two cases of giant malignant eyelid tumors were surgically excised using rapid frozen section margin control. Different pedicle myocutaneous flaps were used to repair the orbital skin defects.

Outcomes: Postoperative follow-up showed perfect healing of the pedicle flaps and good patient compliance. The results of these cases indicate that covering exposed orbital cavities with composite pedicle mycuaneous flaps is a simple and practical strategy for orbital reconstruction. Not only does this help maintain orbital stability, but it also provides opportunities for patients to return to normal lives.

Lessons: Although surgical management is often the first option for treatment of giant eyelid tumors, recurrence and mortality due to the tumors is still high after long-term follow-up. Therefore, early discovery and treatment is the best way to control the progression of giant eyelid tumors and enhance survivability.

Abbreviations: BCC = basal cell carcinoma, CoM = conjunctival melanoma, SCC = squamous cell carcinoma, SGC = sebaceous gland carcinoma.

Keywords: basal cell carcinoma, composite myocutaneous flaps, eyelid tumor, orbit reconstruction, sebaceous gland carcinoma

Editor: Saeed Alzghari.

BF and JJL have contributed equally to this work.

Authors' contributions: LGY designed the study and drafted the manuscript. BF, YJS, and JJL collected and interpreted the data. LGY performed the operation. BFW performed the histopathological examination. All authors read and approved the final manuscript.

Funding/Support: The present study was funded by the National Natural Science Foundation of China (No. 81570864) and the Natural Science Foundation of Jilin Province (No. 20160101004JC; No.20160414045GH; No.2016J041).

Declarations: Written informed consent was obtained from the patients for publication of this case report and any accompanying images. The need for institutional ethics committee approval was waived because the content of this case report did not require ethics approval.

The authors have no conflicts of interest to disclose.

- ^a Department of Ophthalmology, Second Hospital of JiLin University, ChangChun,
 ^b Department of Ophthalmology, First Affiliated Hospital of Harbin Medical University, Harbin, China.
- * Correspondence: Guang-Yu Li, Department of Ophthalmology, Second Hospital of JiLin University, ChangChun 130041, China (e-mail: liguangyu@aliyun.com).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

Medicine (2017) 96:48(e8978)

Received: 27 August 2017 / Received in final form: 7 November 2017 / Accepted: 8 November 2017

http://dx.doi.org/10.1097/MD.0000000000008978

1. Introduction

Giant malignant tumors arising from the eyelid are rare, but often invade various orbital tissues including other parts of the eyelids, eyeballs, lacrimal system, and orbital bones. Thus, extensive orbital skin excision combined with orbital exenteration is necessary in order to reduce tumor recurrence and increase survivability. Because of this, repair of skin defects and reconstruction of the orbital cavity are important issues in surgical management. The current study examines 2 cases of giant malignant eyelid tumors and discusses 2 practical methods for repairing orbital skin defects and maintaining orbital stability.

2. Case 1

A 78-year-old nomad man presented in the Department of Ophthalmology, Second Hospital of JiLin University, with a giant tumor in his right eyelid, which had developed over 3 years.

Clinical examination at admission revealed a huge pigmented, nonhealing ulcerated lesion in the right eyelid. Necrotic tissue was present, along with crusts and scabs in the granulation tissue. The lesion extended superoinferiorly, from the eyebrow to the horizontal level of the nasal alar and mediolaterally, from the nasal dorsum to the lateral canthus (Fig. 1A). On examination, visual acuity was light perception in the right eye and 0.4 in the left eye. Slit lamp examination of the right eye showed tumor invasion into the conjunctiva and sclera accompanied by conjunctival congestion and chromatosis, corneal opacity, and

Fan et al. Medicine (2017) 96:48



Figure 1. Preoperative and postoperative photos of patient with giant eyelid BCC. A. Preoperative photo: double forehead pedicle myocutaneous flaps were designed and transposed. B. Axial view of orbital CT scan showing the tumor engulfing the eyeball and infiltrating the medial lacrimal system (white arrow). C. Postoperative photo at 1-month follow-up: the transposed pedicle flaps had healed perfectly along with the surrounding tissue and the free abdominal flap satisfactorily covered the exposed forehead area. BCC=basal cell carcinoma, CT=computed tomography.

neovascularization. There were no abnormalities observed in the left eye other than lenticular opacity. A computed tomographic (CT) scan of the orbit showed a large skin tumor, approximately $52 \times 44 \times 40 \,\mathrm{mm}^3$ in size, located in the anterior part of the right orbit, engulfing the eyeball, and invading the medial lacrimal system (Fig. 1B). The following differential diagnoses were considered based on these clinical manifestations: basal cell carcinoma (BCC), squamous cell carcinoma (SCC), and conjunctival melanoma (CoM). BCC is the most common type of cancer, constituting 90% of malignant eyelid tumors. Most BCC in Chinese patients is pigmented, however, this subtype is uncommon in white people. [1] BCC is characterized by pearly skin nodules and sometimes presents with ulceration and bleeding. SCC is the second-most common type of skin cancer. The clinical appearance of SCC is highly variable, however, it normally presents as an ulcerated lesion with hard, raised edges, or as a reddish skin plaque that is slow growing and bleeds intermittently. CoM is another common malignant tumor of eye and local tumor seeding can occur spontaneously to the adjacent lid or skin.^[2]

The aim of exenteration is to achieve local control of the disease. Therefore, in the current study, a safe incision margin was made 5 mm outward, along the tumor. The wide surgical excision required removal of all anterior orbital tissue, including the eyeball, lacrimal system, and periorbita along with the eyelids; however, the posterior orbital tissues were preserved. Rapid frozen section margin control was used to guide skin excision.

To reconstruct skin defects after exenteration, double forehead pedicle myocutaneous flaps were transposed to cover the exposed orbital cavity and a free abdominal skin flap was transplanted to cover the exposed area of the forehead. The 1-month postsurgery

follow-up showed that the pedicle flaps, along with the surrounding tissue and skin, had healed perfectly and that the free abdominal flap also healed quickly and satisfactorily (Fig. 1C). The patient was subsequently advised to undergo adjunctive radiotherapy and to undergo follow-up examinations, including orbital CT scans, every 3 months.

Pathological examination revealed that the tumor was BCC. Characteristic epithelial lobules of cells with oval nuclei and scanty cytoplasm with prominent peripheral palisading of the nuclei and elongated strands of basaloid cells embedded in a dense fibrous stroma were observed. The tumor cells had aggressively and deeply infiltrated the adjacent dermis, conjunctiva, sclera and orbital structures (Fig. 2).

3. Case 2

A 52-year-old rural male farmer reported to the Department of Orbital Surgery complaining of a huge tumor in the right eyelid. Patient medical history revealed that the mass was initially the size of a soybean and gradually grew over 3 years to the size of a fist. Vision in the right eye diminished as the tumor became bigger and was completely gone approximately 1 year prior to examination. The patient had delayed proper medical treatment due to financial difficulties.

Upon physical examination, the basal part of the tumor was found to be derived from right lower eyelid and was growing eccentrically. The tumor invaded the right inner canthus and lacrimal system medially, reached the right side of maxillary buttress laterally and extended from supraorbital ridge to the horizontal level of nasal alar superoinferiorly. The tumor surface was approximately $7 \times 7 \times 8 \, \mathrm{cm}^3$, ulcerative, nodular, exophytic, and had purulent blood exudate. The skin of the upper eyelid

Figure 2. The excised tumor sample and histopathological photo. A. Pigmented nodular and noduloulcerative lesion approximately $52 \times 44 \times 40 \,\mathrm{mm}^3$ in size. B. Histopathology pictures: nodules of epithelial tumor cells lined by peripheral palisading of the nuclei (HE staining, scale bar = $100 \,\mu\mathrm{m}$).

appeared to be intact. The vision in the left eye was normal, ocular movements in all directions were intact and all other parameters were within normal range. The axial and coronal sections of the orbital CT showed that the eyeball was squeezed superiorly and laterally, that the lesion occupied most of the orbital cavity space and that the orbital cavity was extruded and increased; however, no obvious erosive damage was observed in the bony structures (Fig. 3). Based on the clinical history, examination, and radiological findings, a provisional diagnosis of carcinoma of right lower eyelid was suggested and sebaceous gland carcinoma (SGC) was highly suspected considering it was a solitary nodule with an eccentric growth pattern.

A safe surgical excision (5 mm skin around the tumor base including the lacrimal system and lateral canthus) was performed and skin margin control was inspected with rapid frozen sections. The orbital contents were completely enucleated and a canal connecting the orbital cavity to the ethmoid sinus was made in order to drain the cavity exudatives. The normal skin and subcutaneous muscle layer of the upper eyelid was preserved

(Fig. 4A). Finally, a pedicle myocutaneous flap from the upper eyelid was transposed inferiorly to cover the orbital cavity and sutured with a skin incision margin. Histopathological examination revealed SGC with necrosis in the center of the tumor. The tumor contained large anaplastic cells that exhibited sebaceous differentiation, hyperchromatic nuclei, and characteristic features of vacuolated foamy-frothy cytoplasm (Fig. 5).

At the 3-week follow up, the flap had healed satisfactorily and the patient had good compliance (Fig. 4B–D). The patient was advised to undergo further adjunctive radiotherapy and to undergo follow-up orbital examinations and general physical checkups every 3 months to reduce the risk of recurrence.

4. Discussion

Giant eyelid tumor is a descriptive diagnosis, which indicates a tumor that is relatively huge in size and/or has an extensive invasion range. The formation of a giant eyelid tumor depends on the nature of the tumor, the growth rate, and duration of the

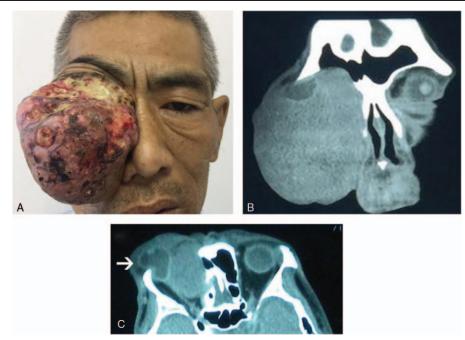


Figure 3. Preoperative photo of the patient with giant eyelid BCC and orbital CT scan. A. Giant eyelid-derived tumor approximately $7 \times 7 \times 8 \,\mathrm{cm}^3$ in size; B, coronal view, and C. axial view of CT scan: the lesion invaded orbital content and the eyeball was superiorly and laterally squeezed (white arrow). BCC=basal cell carcinoma, CT=computed tomography.

Fan et al. Medicine (2017) 96:48

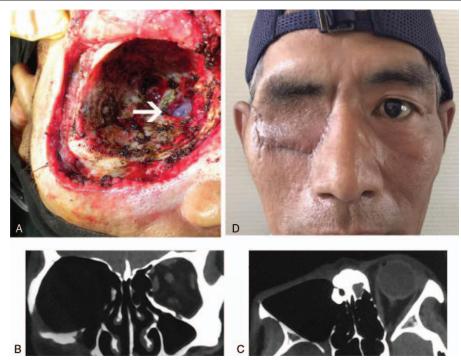


Figure 4. Surgical image. Postoperative CT images and postoperative photo of patient. A. The complete orbital content was enucleated and a canal between the orbital cave and enthmoid sinus was established (white arrow); B, C. Postoperative CT view shows that an empty orbital cavity without effusion is covered with a myocutaneous flap; D. Postoperative photo of patient at 3-week follow-up. CT=computed tomography.

tumor. Giant eyelid tumors are also related to the educational level and financial condition of patients, as well as to access to local health care. Based on clinical observations, most giant eyelid tumors are malignant and commonly include basal cell carcinoma, cutaneous squamous cell carcinoma, sebaceous gland carcinoma, and cutaneous melanoma. [3–7] However, some giant eyelid tumors are benign, such as neurofibromatosis, [8] pilomatrixoma, [9] and xanthogranuloma. [10]

The primary therapeutic principles for treating giant malignant eye tumors are to increase survivability and avoid tumor recurrence. Therefore, surgical removal should be performed as quickly as possible using frozen section control in order to achieve safe incision margins. Cosmetic considerations can be thought of as secondary therapeutic principles and are often based on the age and specific requirements of each patient. Once

the tumor infiltrates the eyeball or orbital contents, anterior orbital exenteration, or complete orbital exenteration must be done. To avoid the orbital cavity effusion, a canal between orbital cavity and ethmoid sinus can be established during the surgery to drain the orbital exudates. Whether the lacrimal system and intact upper or lower eyelid, or a part of myocutaneous tissue of the eyelids are preserved, should be considered preoperatively, based on the surgical excision scale. The surgical excision scale is a balance between the clear excision of tumor-infiltrated tissues and the preservation of healthy skin for use in reconstruction. Certainly, if the tumor invades orbital bones or local lymph node metastasis occurs, more extensive surgical excision should be planned, including removal of the infiltrated orbital bones and local lymph node dissection (antero auricular lymph nodes and superficial anterior cervical lymph node), cooperatively with oral

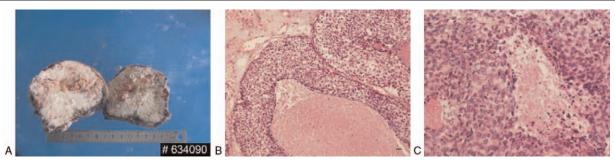


Figure 5. The excised tumor sample and histopathological photo. A. The tumor with necrosis in the center, approximately $7 \times 7 \times 8$ cm³ in size. B. Histopathology pictures: sebaceous gland carcinoma (SGC), the tumor contained large anaplastic cells that exhibited sebaceous differentiation, showed hyperchromatic nuclei and characteristic features of vacuolated foamy-frothy cytoplasm (HE staining, scale bar = $25 \mu m$).

surgeons. After excision, the orbital cavity must be reconstructed to maintain the stability of the orbit and to create a relatively acceptable appearance so that patients can resume normal lives. Therefore, composite myocutaneous flaps are normally used to cover the exposed orbital cavity. Since there is no blood supply under the transposed flaps, pedicle myocutaneous flaps are the best choice. These may come from forehead, healthy eyelid, or cheek. Defects of the forehead skin can be repaired using a free abdominal flap. Additionally, one-stage reconstruction of the orbital cave, using a transposed temporalis muscle flap, should be carefully considered since the recurrence tumors is uncertain and postoperative radiotherapy may influence temporalis muscle flap survival. Repairing orbit defects with pedicle myocutaneous flaps is a simple and practical method. However, the limitation of this surgical procedure is that orbital rehabilitation is not taken into consideration and patients may need further operations to install orbital implants in order to gain better cosmetic results.

Giant eyelid tumors are highly associated with low socioeconomic status. It is strongly advised that medical insurance should extensively cover people in economically deprived areas and that people living in those areas should be advised to have regular physical examinations. Early discovery and treatment is the best way to control the formation of giant eyelid tumors and to increase survivability. As evidenced by these 2 cases, completing surgery is not enough; it is vital that potential social factors

associated with giant eyelid tumors be taken into account. Therefore, we are trying to enroll the patients from this study in a program sponsored by Red Cross Society of China that provides assistance funding to relieve the financial burden of medical follow-ups for these patients.

References

- [1] Margo CE, Waltz K. Basal cell carcinoma of the eyelid and periocular skin. Surv Ophthalmol 1993;38:169–92.
- [2] Pe'er J. Pathology of eyelid tumors. Indian J Ophthalmol 2016; 64:177–90.
- [3] Moussala M, Behar-Cohen F, D'Hermies F, et al. [Giant basal cell carcinoma of the eyelid in a black patient from Cameroon]. J Fr Ophtalmol 2000;23:595–8.
- [4] Fetohi M, Mazghi AE. Giant basal cell carcinoma of the eyelid: a case history. Pan Afr Med J 2016;24:281.
- [5] Wali UK, Al-Mujaini A. Sebaceous gland carcinoma of the eyelid. Oman J Ophthalmol 2010;3:117–21.
- [6] Weiling M, Bergua A, Kruse FE, et al. [Therapy options for malignant eyelid tumors]. Ophthalmologe 2016;113:1095–108.
- [7] Silverman N, Shinder R. What's new in eyelid tumors. Asia Pac J Ophthalmol (Phila) 2017;6:143-52.
- [8] D'Hermies F, Hurbli T, Morel X, et al. [Eyelid neurofibroma affecting a young woman]. J Fr Ophtalmol 2002;25:333–6.
- [9] Levy J, Ilsar M, Deckel Y, et al. Eyelid pilomatrixoma: a description of 16 cases and a review of the literature. Surv Ophthalmol 2008;53:526–35.
- [10] Vick VL, Wilson MW, Fleming JC, et al. Orbital and eyelid manifestations of xanthogranulomatous diseases. Orbit 2006;25:221–5.