• Brief Report •

Optimizing surgical approaches for lacrimal gland adenoid cystic carcinoma to minimize cross-organ invasion

Ming-Shen Ma[1,3], *Ting-Ting Ren*[2], *Fu-Xiao Luan*[1], *Jing Li*[2], *Nan Wang*[2], *Yong Tao*[1], *Jian-Min Ma*[2]

[1]Department of Ophthalmology, Beijing Chaoyang Hospital, Capital Medical University, Beijing 100027, China
[2]Department of Ocular Oncology, Beijing Tongren Hospital, Capital Medical University, Beijing 100005, China
[3]Gies College of Business, University of Illinois at Urbana-Champaign, Champaign, IL 61820, United States
Co-first authors: Ming-Shen Ma and Ting-Ting Ren
Correspondence to: Jian-Min Ma. Department of Ocular Oncology, Beijing Tongren Hospital, Capital Medical University, Beijing 100730, China. jmma@ccmu.edu.cn; Yong Tao. Department of Ophthalmology, Beijing Chaoyang Hospital, Capital Medical University, Beijing 100020, China. taoyong@mail.ccmu.edu.cn.
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Abstract

● **AIM:** To evaluate the outcomes of eye-sparing surgery for lacrimal gland adenoid cystic carcinoma and the impact on tumor recurrence and orbital integrity.

● **METHODS:** The study enrolled four patients with recurrent lacrimal gland adenoid cystic carcinoma. The outcome focused on the relevance of the integrity of the lateral orbital wall to the occurrence of extraorbital metastasis in the local recurrence of lacrimal gland adenoid cystic carcinoma.

● **RESULTS:** Three patients underwent eye-sparing surgery *via* lateral orbitotomy without postoperative radiotherapy, and one patient who underwent eye-sparing surgery *via* sub-brow approach. These four patients all demonstrated a recurrence involving the invasion of extraorbital tissues as metastatic form through surgical bone seams.

● **CONCLUSION:** Preserving intact orbital bone tissue is crucial for mitigating direct cross-organ metastasis of lacrimal gland adenoid cystic carcinoma. The findings suggest avoiding the lateral orbitotomy approach with no or limited orbital bone wall invasion.

● **KEYWORDS:** lacrimal gland adenoid cystic carcinoma; recurrence; eye-sparing surgery
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INTRODUCTION

Lacrimal gland adenoid cystic carcinoma (LGACC) is the most common primary malignant epithelial tumor of the lacrimal gland[1]. Despite extensive research, the pathogenesis of LGACC remains unclear, and it is characterized by high rates of recurrence, metastasis, and mortality, posing significant clinical challenges[2]. Surgical intervention, guided by the American Joint Committee on Cancer TNM staging system for LGACC, remains the primary treatment modality[3]. For early to mid-stage LGACC, eye-sparing surgery followed by postoperative radiotherapy is the standard approach[4]. However, the surgical techniques and approach selections that potentially influence postoperative recurrence and prevent extraorbital organ invasion relations are still unclear.

SUBJECTS AND METHODS

Ethical Approval The study adhered to the principles of the Declaration of Helsinki and was approved by the Ethics Committee of Beijing Tongren Hospital, Capital Medical University (TRECKY2019-093). The written consent forms have been collected.

The study enrolled four patients with recurrent LGACC who underwent lateral orbitotomy without postoperative radiotherapy at Beijing Tongren Hospital, Capital Medical University. Basic information collected from patients included age, sex, clinical manifestations, treatment, and prognosis. The outcome focused on the relevance of the integrity of the lateral orbital wall to the occurrence of extraorbital metastasis in the local recurrence of LGACC evaluated by imaging studies [magnetic resonance imaging (MRI) or computed tomography (CT)] and histopathological examination.

RESULTS

Case 1 A 52-year-old male underwent lateral orbitotomy one year ago to remove a tumor in the left orbit. Postoperative

pathology confirmed poorly differentiated LGACC. He received traditional Chinese medicine for eleven months post-surgery, without undergoing radiation therapy. Upon re-examination at our hospital, MRI and CT scans revealed recurrence, with the lesion involving the lateral orbital wall and extending to the subcutaneous layer (Figure 1A, 1B). Intraoperative findings showed visible extraorbital lesion tissues (Figure 1C, 1D).

Case 2 A 38-year-old male presented with a five-month history of right eye proptosis and underwent lateral orbitotomy for tumor excision one month prior. Postoperative pathology confirmed solid-type LGACC. He did not receive postoperative radiation therapy. Follow-up MRI revealed recurrence of the lesion in the right orbit, extending to the orbital apex and metastasizing to the subcutaneous temporal fossa of the lateral orbital wall (Figure 2).

Case 3 A 33-year-old female presented with left eyelid swelling ten months ago. Imaging suggested an occupying lesion in the left orbit, and she underwent lateral orbitotomy for tumor excision. Postoperative pathology confirmed LGACC. She received three courses of chemotherapy. Follow-up MRI and CT scans showed a left intraorbital lesion, with involvement of the subcutaneous tissue and temporal muscle outside the orbit. The bone seam created by the surgery was visible (Figure 3).

Case 4 A 58-year-old female patient underwent eye-sparing surgery for left orbital LGACC *via* the sub-brow approach. Eleven years later, follow-up orbital MRI and CT scans revealed a postoperative recurrence (Figure 4). The recurrent lesion was confined within the orbit, with no evidence of direct extension beyond the orbital boundaries. Consequently, we performed a second eye-sparing surgery.

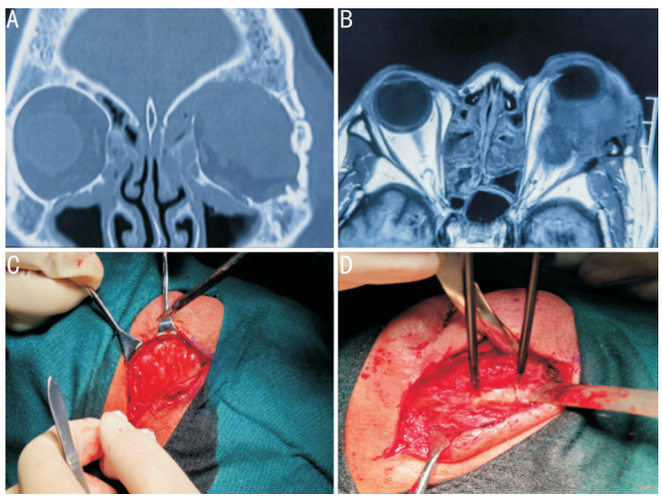

Figure 1 Imaging of extraorbital metastatic lesions in Case 1 A, B: A large solid occupying lesion visible inside and outside the left orbit, including the bone seam caused by surgery on the lateral orbital wall; C, D: The incision of subcutaneous tissue, exposing metastatic lesions migrating from the orbit through the lateral wall, with the bone flap seam visible after removing the affected tissue.

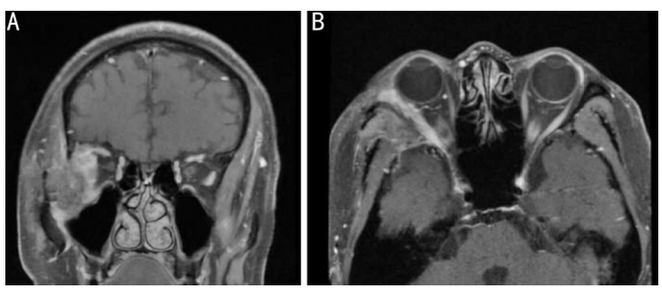

Figure 2 The MRI images showed a lesion within the right orbit with involvement at the temporal fossa (A) extending from the lacrimal gland area to the orbital apex (B).

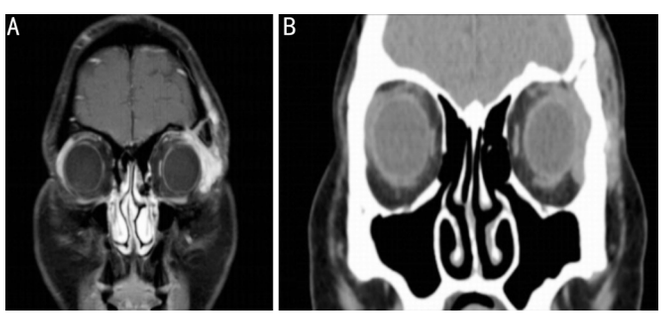

Figure 3 The MRI (A) and CT (B) scans showed a lesion inside the left orbit with involvement of the extraorbital subcutaneous tissue and temporal muscle, clearly displaying the bone seam caused by surgery and direct invasion of extraorbital tissues by recurrent tumors through the bone seam.

DISCUSSION

Previous studies have primarily focused on the choice between orbital exenteration and eye-sparing surgery, and the subsequent decisions regarding adjuvant radiotherapy or chemoradiation[1-2,5]. These studies often overlook the surgical approach in eye-sparing surgeries for LGACC. Our clinical experience indicates that the surgical approach significantly impacts the prognosis of LGACC patients.

For early to mid-stage LGACC, eye-sparing surgery is generally preferred to preserve visual function and appearance[4]. However, there is no standardized surgical approach for eye-sparing surgery in LGACC. It remains unclear whether to use the sub-brow orbital rim incision or the lateral orbitotomy approach. The lateral orbitotomy approach, a standard method for orbital tumor surgeries, provides a broader surgical field of view but involves cutting through the lateral orbital bone walls. Although the bone walls are typically repositioned postoperatively with materials such as titanium alloys or absorbable substances, the bone seams created during surgery often become pathways for tumor recurrence, leading to extraorbital tissue invasion and metastasis.

In this study, three patients had undergone lateral orbitotomy surgeries without receiving postoperative local radiation therapy. Each experienced extraorbital invasion of LGACC,

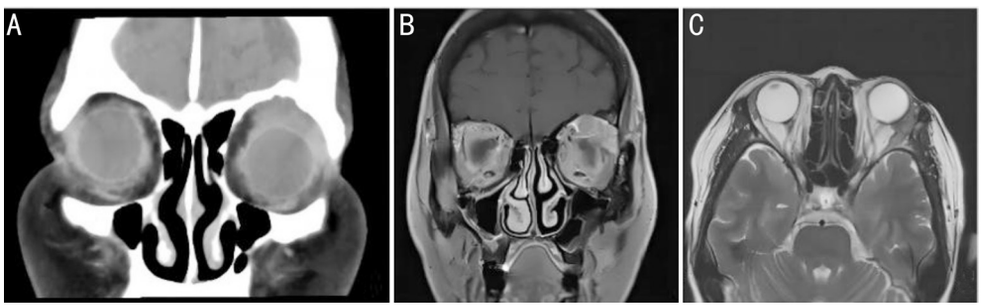

Figure 4 The CT scan images demonstrated that the orbital bony walls were continuous (A) and MRI scan images showed that the recurrent lesion was confined within the orbit (B, C).

with imaging suggesting that the invasion was related to the bone flap gap created by the lateral orbitotomy approach (Figure 1C, 1D). During secondary surgeries, significant tumor tissue migration around the lateral orbital wall bone seam was observed, demonstrating that opening the lateral wall can lead to extraorbital invasion. On the other hand, in one patient who had undergone a sub-brow orbital rim incision, the postoperative recurrence was confined within the orbit.

During the surgery for orbital malignant tumors, the orbital bone needs to be reconstructed. The gaps caused by the surgery or tumor invasion may become channels for orbital malignant tumors to invade extraorbital tissues. Incomplete excision can leave residual cells that may lead to recurrence. Various implant materials, such as hydroxyapatite, titanium meshes, bioactive glass, and 3D-printed implants, were used for orbital reconstruction and have a long history of development in this field[6-12].

However, most of the current and traditional implants are primarily designed to reconstruct orbital fractures, and they do not provide a physical barrier to prevent malignant tumor cells from invading the orbital bone tunnels. In a previous case report, researchers proposed the modified lateral orbitotomy Approach, using titanium materials to repair the bone sutures, and suggested it as an effective surgical method for treating LGACC[13]. While this approach showed promise, the study had limitations, including a small sample size and the absence of a control group. Given these considerations, it is essential to prioritize a surgical approach that avoids destruction of the orbital bone wall. Additionally, compared to patients treated *via* the sub-brow approach, those who underwent the modified lateral orbitotomy approach or lateral orbitotomy may have a higher risk of significant postoperative facial scarring. Therefore, careful selection of the surgical technique is critical to balancing oncological safety and aesthetic outcomes.

Previous studies have focused on the choice between orbital exenteration and eye-sparing surgery, often overlooking the surgical approach in eye-sparing surgeries for LGACC. Our study highlights the significance of the surgical approach in influencing prognosis. The lateral orbitotomy approach, despite providing a broader field of view, creates bone seams that serve as pathways for tumor recurrence and metastasis. In contrast, the sub-brow orbital rim incision preserves the integrity of the orbital bone, reducing the risk of extraorbital invasion. Therefore, we recommend using the sub-brow orbital rim incision in cases with no or limited orbital bone wall invasion to preserve the integrity of the orbital bone tissue and prevent extraorbital tissue invasion upon recurrence. This approach reduces the likelihood of intraorbital recurrence directly invading extraorbital tissues, enabling secondary intraorbital tumor resections and preventing disfigurement and life-threatening conditions from an expanded range of tumor recurrence. In this clinical case series study, we compared three cases of recurrent LGACC treated *via* lateral orbitotomy with one case treated *via* the sub-brow approach. Due to the small sample size and the inherent limitations of individual clinical cases, we cannot draw a definitive conclusion regarding the suitability of lateral orbitotomy for LGACC removal, particularly when complete and safe tumor resection may not always be achievable through the sub-brow approach. Postoperative local radiation therapy can be administered to destroy any remaining tumor cells. Future research should focus on refining these surgical techniques and exploring artificial material to fix the bone tunnels directly or indirectly induced by LGACC to further improve patient outcomes.

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