CASE REPORT

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# Reconstruction of a Misleading Sebaceous Gland Lesion of an Eyelid: A Case Report

## Göz Kapağında Yer Alan Yanıltıcı Bir Sebase Gland Lezyonunun Rekonstrüksiyonu: Olgu Sunumu

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## Abstract

Sebaceous carcinoma is a highly malignant eyelid tumor with a notable morbidity and mortality rate. Delay in the diagnosis occurs usually; due to benign formations that are confused in the differential diagnosis. We report the case of sebaceous gland carcinoma in a 61-year-old male patient who presented with a long-standing nodule on the right upper eyelid and had right parotid lymph nodes metastases. After wide excision, reconstruction of the oncologic defect was performed considering the functional and aesthetic results. No residual disease was found in the 4-year follow-up. Successful management of sebaceous gland carcinoma consists of early diagnosis, complete removal of the tumor, and a satisfactory reconstruction method. Treatment should be designed on the basis of the tumor and the patient's needs.

**Keywords:** Eyelid, reconstruction, sebaceous carcinoma, wide local excision

## Öz

Sebase karsinom, önemli bir morbidite ve mortalite oranına sahip oldukça malign bir göz kapağı tümörüdür. Ayırıcı tanıda benign lezyonlar ile karışmasından dolayı tanıda gecikme sık yaşanır. Sağ üst göz kapağında uzun süredir var olan nodül ve sağ parotis lenf nodu metastazı ile başvuran 61 yaşında erkek hastada tanı konan sebase bez karsinomu sunulmaktadır. Geniş eksizyon sonrası fonksiyonel ve estetik sonuçlar göz önünde bulundurularak onkolojik defekt rekonstrüksiyonu yapılmıştır. Dört yıllık takibinde rezidüel hastalık saptanmamıştır. Sebase gland karsinomunun başarılı tedavisi, erken teşhis, tümörün tamamen çıkarılması ve tatmin edici bir rekonstrüksiyon yönteminden oluşmaktadır. Tedavi, tümöre ve hastanın ihtiyaçlarına göre tasarlanmalıdır.

**Anahtar kelimeler:** Geniş lokal eksizyon, göz kapağı, rekonstrüksiyon, sebase karsinom

## Introduction

The eyelids are an anatomical region where roughly 5-10% of all skin cancers occur (1). The most frequent skin tumors in this region are basal cell carcinoma (BCC) and squamous cell carcinoma (SCC) (2). Sebaceous gland carcinoma (SGC) is a rare tumor with morbidities, such as up to orbital exenteration and mortality. This tumor can develop in Meibomian, Zeis, and other sebaceous glands of the eyelid (3). In 1891, Allaire described SGC for the first time (4). A rare cancer that primarily affects the head and neck region. It can behave aggressively and can be either ocular or

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extraocular (5). The ocular region is seen more commonly (34.5-59%) (6), and according to a study by Orr et al. (5), the eyelids are involved in 39% of SGCs. Tumors in the ocular region are seen mainly in the Meibomian, caruncle, and Zeiss glands and eyebrows. Upper eyelids are more frequently affected than lower eyelids because they contain more sebaceous glands. The average age at diagnosis was 67 years. Tumors are more common in women (7). Demographically, Asian people are affected more than other ethnic groups by these tumors (8).

SGC can mimic benign lesions. Therefore, delayed diagnosis is frequently observed. Initially, a preliminary diagnosis can be made of chalazion, chronic conjunctivitis, posterior blepharitis, or Meibomian cyst. In the advanced stages, when the lesion resists local treatment or surgical drainage, the preliminary diagnosis is SCC, BCC, or even lymphoma (9).

The clinical manifestations of madarosis include destruction of the eyelid edge, generalized thickening of the eyelid, and facial shrinkage (10). In the present case, the telangiectatic appearance in the anterior part of the lesion suggests BCC; the fleshy, degradable lower part of the lesion suggested SCC.

Pathogenesis is unknown. Predisposing factors include Muir-Torre syndrome (MTS), periocular radiation, hereditary retinoblastoma, and immunosuppression (10). MTS is an inherited autosomal dominant syndrome linked to sebaceous adenoma or carcinoma, as well as gastrointestinal, endometrial, and urological malignancies. Therefore, patients with SGC should be aware of long-term internal malignancies. Although the patient did not have any radiation exposure or other comorbidities in his/her history, SGC was diagnosed.

SGC has three histological growth patterns: Lobular, comedocarcinoma, and papillary carcinoma. The presented patient's histological type is lobular carcinoma, the most common type (11). The different morphological patterns in the lesion may overlap. Some immunohistochemical staining and proteins can be used for the diagnosis of SGC. Of these, EMA, Ber-Ep4, androgen receptor, and adipophilin were found to be significant (11). EMA, Ber-Ep4, and adipophilin protein were positive in the present case. In lobular patterns, poorly differentiated cells are peripheral between normal sebaceous glands; well-differentiated lipid-producing cells are located in the center. The papillary pattern typically presents as a small conjunctival tumor with fleshy papillary projections. The Comedocarcinoma subtype is characterized by a large central necrotic core and living cells around it (12).

Poor prognostic factors include longer than six months, vascular-lymphatic infiltration, poor differentiation, extension to the orbit, pagetoid spread, intraepithelial carcinomatous alteration, and upper eyelid involvement. In the present case, a nodular mass was observed on the upper eyelid for 5 years, suggesting poor prognosis. Recurrence occurs in 9-36% of cases. Local metastasis occurs in the parotid, submandibular, and cervical lymph nodes. At the same time, distant metastases occur in the lung, liver, skull, and brain (13), according to a study by Sa et al. SGC with a diameter >20 mm was correlated with an increased risk of local recurrence, lymph node metastasis, distant metastasis, and death from the disease (7). When a tumor involves the entire eyelid, the risk of lymph node and distant metastases increases, even if the tumor diameter exceeds 10 mm. In the study, local recurrence was 6%, lymph node metastasis was 21%, distant metastasis was 7%, and the mortality rate from the disease was 6%. After definitive treatment, metastasis and local recurrence are usually observed within the first 2 years. Metastasis after 5 years is extremely rare (7).

In such an important facial esthetic unit, surgery is the mainstay of treatment. Total parotidectomy, radical neck dissection, and radiation therapy are recommended for patients with regional lymph node metastases. Cryotherapy or radiotherapy may be beneficial for patients who are not candidates for surgery (10). Chemotherapy is used for recurrent, metastatic lesions, but there is no high-level of evidence (13).

This report aimed to present the excision and reconstruction of a lesion that mimics a benign lesion that has been present for a long-time in an esthetically and functionally significant facial unit.

## Case Report

A 61-year-old male patient was admitted to the plastic surgery outpatient clinic with the complaint of a mass on the right upper eyelid that had developed in 5 years. Initially, the patient was misdiagnosed because the palpebral mass was negligible and confused with infection. On clinical examination, a firm, painless nodule-like mass was observed covering more than 80% of the right upper eyelid (Figure 1A). Except for hypertension and long-term smoking, the patient had no additional disease and no characteristics-feature in his family history.

The patient underwent magnetic resonance imaging (MRI), ultrasonography, and positron emission tomography for tumor characterization. Orbital MRI did not reveal any expansion into the orbit. In addition, no pathological lymph nodes were detected on neck ultrasonography. Positron emission tomography revealed 11-mm hypermetabolic metastatic lymphadenopathy in the right preauricular region and a 2-cm hypermetabolic area in the right parotid gland.

Tru-cut biopsy was performed on the parotid gland. The presence of carcinoma infiltration was confirmed. According to the American Joint Committee, the patient had regional cancer of grade T(3c)N(Ia)M(0) that had spread only to the nearby right preauricular lymph nodes and right parotid gland. The ophthalmology department did not recommend orbital excision. Two-stage treatment was administered in March 2018. In the first operation, the tumor was completely excised, with a full-thickness upper eyelid and a 5-mm margin of intact skin (Figure 1B). After resection, a 3.5 cm x2 cm full-thickness tissue defect affecting 80% of the upper eyelid was formed (Figure 1C). The posterior lamella of this defect was reconstructed with a transconjunctival interpolation flap, similar to the Cutler beard flap. The anterior lamella was reconstructed using a lower eyelid-based fasciocutaneous transposition flap. The right lower eyelid donor site was reconstructed using a right cheek advancement flap (Figure 1D). Simultaneously, superficial parotidectomy was performed. The pathology department diagnosed the patient with SGC. After the patient's first surgery, six sessions of adjuvant chemotherapy were administered to the oncology clinic.

In the second operation, after waiting for revascularization, the interpolation flap was separated (Figure 2). Later, the right upper eyelid was revised due to lateral canthal narrowing. The right cheek advancement flap was removed for attachment to the lateral canthus. A conjunctival flap is applied to complete the periorbital reconstruction.

After discharge, follow-up was performed at 3-month intervals in the plastic surgery and oncology outpatient clinics.

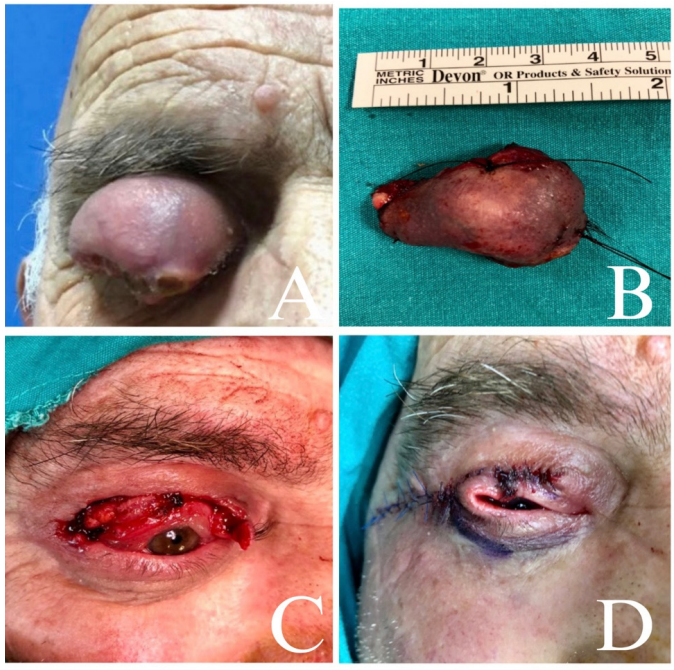


**Figure 1. A.** Preoperative view of the mass covering 80% of the right upper eyelid, **B.** 3.5 cm x2 cm sized excisional biopsy material, **C.** The right upper eyelid defect, **D.** First session: tarsoconjunctival interpolation flap and cheek advancement flap

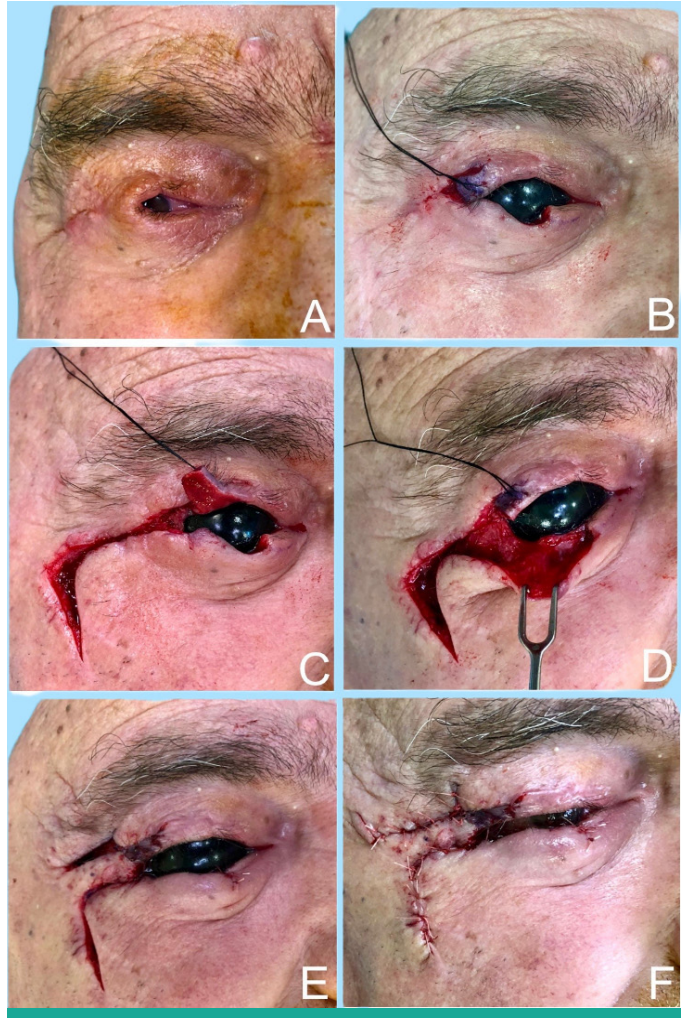


**Figure 2. A.** Lateral canthal narrowing, **B.** Interpolation flap is shown, C. Interpolation flap separation, **D-F.** Advancement of right cheek advancement flapand upper eyelid flap to lateral canthus.

In early and late follow-ups, muscle integrity was not impaired while blinking. There is no dry eye or lagophthalmos. The second and fourth-year follow-up images demonstrate bilateral symmetry (Figure 3) without ectropion, contracture, or lagophthalmos. No complications or recurrent lesions were observed during follow-up. The oncology council recommended radiation therapy as an adjuvant therapy.

This study is a case report, and the University of Health Sciences Turkey Ethics Committee confirmed that there was no need for ethics committee approval. The patient provided informed consent. Research related to human use was conducted in accordance with the tenets of the Helsinki Declaration.

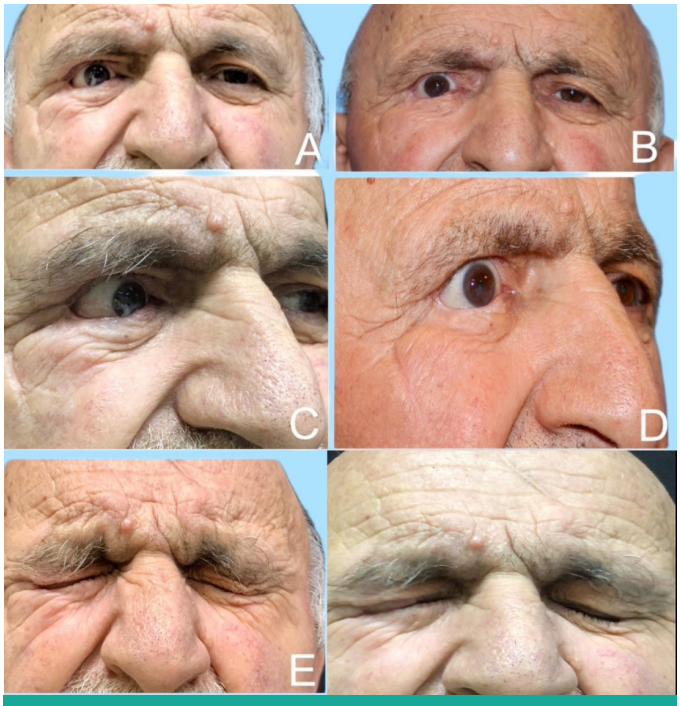


**Figure 3.** The patient's dynamic pictures in the postoperative second **(A, C, E)** and forth **(B, D, F)** years demonstrate minimal deformity with optimal aesthetic and functional results

## Discussion

The eyelids form a major esthetic and functional unit of the upper face. Trauma and oncologic resection are common etiologies of upper face defects. In the present case, the initial specimen obtained from the right upper eyelid tumor 5 years previously corresponded to an infected lesion. Diagnosis was delayed and misdiagnosed because of poor sebaceous differentiation.

Treatment is shaped according to the histopathological type, stage, and patient preference. However, surgery is a cornerstone in the treatment planning. After full-thickness eyelid biopsy, a wide excision with a safe surgical margin of 5 mm is recommended. When the tumor involves the bulbar conjunctiva and periorbital reconstruction is not possible, exenteration is required. A well-designed reconstruction should be designed to cover the defect after oncological resection to restore eyelid function and achieve a satisfying esthetic result. In patients with full-thickness eyelid defects, anterior and posterior lamella reconstruction should be performed. Although there are various algorithms for reconstructing eyelid skin and subcutaneous tissue (Table 1), planning is performed individually and requires the surgeon's creativity; according to the current literature, reconstruction options are still limited for total or near total upper eyelid defects. Defects occurred after excision of SGC with healthy margins that required appropriate posterior and anterior lamellar reconstruction. In similar cases, some eyelid-sharing methods, such as Cutler beard flap, Switch flap, or pericranial galeal flap, or Tenzel's semicircular flap, can be preferred (14-16). Eyelid-sharif methods inherently pose patient discomfort and temporary visual impairment but provide "like for like" reconstruction options. Other flap options, such as cheek advancement or Tenzel's semicircular flaps, have less tissue, similar to periorbital flap options. In the present case, transconjunctival interpolation and fasciocutaneous local flap were devised because of the local flap's advantages. These include tissue, color, and shape harmony. As a result, the eyelid function was improved.

**Table 1. Algorithm for full-thickness upper eyelid reconstruction based on the defect size (14)**

| Size of eyelid margin defect (eyelid width) | Repair |
|---|---|
| <25% | Direct closure |
| 25-50% | Direct closure with lateral cantholysis |
| 25-50% | Tarsal rotation flap and skin-muscle flap or skin graft |
| 25-75% | Tarsoconjunctival graft and skin-muscle flap |
| 33-66% | Semicircular flap with periosteal flap |
| 50-100% | Cutler-beard flap |

## Conclusion

SGC accounts for less than 1% of all skin malignancies. Diagnosis may be delayed for years due to its tendency to mimic benign eyelid lesions. To prevent high morbidity and mortality in patients with this cancer, clinicians should always be cautious in the preliminary diagnosis. Treatment methods vary from one patient to another, according to the histopathological characteristics of the tumor and its dissemination. However, wide excision and functionally, esthetically acceptable reconstruction are the basis of this treatment.

### Ethics

**Informed Consent:** The patient provided informed consent.

### Authorship Contributions

Surgical and Medical Practices: P.K., L.J.M.S., S.Ö., K.Ö., Concept: L.J.M.S., S.Ö., K.Ö., Design: L.J.M.S., S.Ö., K.Ö., Data Collection or Processing: P.K., L.J.M.S., S.Ö., K.Ö., Analysis or Interpretation: P.K., S.Ö., K.Ö., Literature Search: P.K., S.Ö., Writing: P.K., L.J.M.S., S.Ö., K.Ö.

**Conflict of Interest:** No conflict of interest was declared by the authors.

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