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A case of sebaceous carcinoma in the eyelid: The results of chemoradiotherapy after recurrence

Göz kapağında sebase karsinom olgusu: Nüks sonrası kemoradyoterapi sonuçları

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Abstract
Sebaceous carcinoma of the eyelid, a rare and locally aggressive malignant neoplasm, can metastasize via the lymphatic and hematogenous route. The diagnosis and treatment can be delayed because of the similarity of the tumors with benign lesions. We herein present a 65-year-old male patient who was diagnosed with sebaceous carcinoma of the left upper eyelid 7 years ago, after which surgical resection was performed. Regional recurrence developed in left cervical nodes two years later. Left neck dissection was performed and adjuvant radiotherapy was administered. The patient has no evidence of recurrence or metastasis after radiotherapy during 5 years of follow-up.
Keywords: Sebaceous carcinoma, Radiotherapy, Eyelid

Öz
Göz kapağının sebase karsinomu nadir ve lokal-agresif malign neoplazmıdır. Lenfatik ve hematojen metastaz görülebilir. Benign lezyonlara benzerliği nedeniyle tanı ve tedavi gecikebilir. Bu olgu, 7 yıl önce sol üst göz kapağında sebase karsinom tanısı konulan 65 yaşında erkek hastadır. Cerrahi rezeksiyon yapıldıktan 2 yıl sonra sol servikal lenf nodunda nüks gelişti. Sol boyun diseksiyonu ve daha sonra adjuvan radyoterapi uygulandı. Hastanın 5 yıllık gözlem sırasında radyoterapiden sonra nüks veya metastaz bulgusu yoktur.
Anahtar kelimeler: Sebase karsinom, Radyoterapi, Göz kapağı

Introduction

Sebaceous carcinoma (SC) arises from the sebaceous gland of the skin for which there is no standard treatment. In general, it occurs in the periorbital and head and neck regions (extraocular). Due to similarities with benign lesions, it can be misdiagnosed. Early treatment is significant for locoregional control. Lymph node metastasis is common (30%) [1]. We aimed to share the results of a case in which multiple treatment modalities were applied.

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Case presentation

A 65-year-old male presented with complaint of a mass in the left upper eyelid. It was decided to evaluate the orbital region with contrast-enhanced MRI for preliminary diagnosis, which revealed a 24x18x10 mm heterogeneous hyperintense lesion in the left upper eyelid. Ultrasonographic evaluation of the cervical region was negative for any metastatic lymph nodes. Local excision was performed without sentinel lymph node biopsy (SLNB) or lymph node dissection. Pathological diagnosis was sebaceous carcinoma with a 1 mm surgical margin and a tumor size of 22 mm. After 2 years, the patient presented with left upper cervical mass. Relapse was considered and ^{18}F fluorodeoxyglucose positron emission tomography/computed to mography (FDG PET/CT) was performed for re-staging, which showed left upper jugular (12 mm) (SUVmax=14) and left submandibular (23x16 mm) (SUVmax=14) hypermetabolic lymphadenopathies. The patient then underwent left selective neck dissection. The pathology report showed a total of 5 lymph node metastases, 4 of which were level I and 1 of which was level II. After surgery, radiotherapy (RT) was administered along with weekly cisplatin treatment. EBRT was delivered to the left level I-IV, preauricular and intraparotid lymph node regions with 6MV X-rays using Intensity Modulated Radiotherapy (IMRT). The patient was treated with a median radiation dose of 60 Gy to level I-II nodal regions and 54 Gy to elective nodal regions (level III-IV, preauricular and intraparotid lymph nodes) in 2 Gy fraction doses.

He was followed-up at 3-6 months intervals for 5 years after RT. During follow up, there was no local regional progression or distant metastasis.

Discussion

Ocular SC is a rare tumor with poor prognosis arising from the Meibomian glands of the upper eyelid. The incidence varies among ethnic groups. Although it is exceedingly rare in the Western world, its incidence is quite high in China, Japan, India.

It is emphasized in the literature that these cases can be overlooked due to misdiagnosis. In a study by Lam SC et al. [2], it was noted that the delay of treatment had a negative effect on DFS. In this study, 5-year survival rates were better in cases with a disease history of less than 6 months. Our patient was followed up after being misdiagnosed as hordeolum for more than 6 months until the correct diagnosis.

SC is common in older female individuals, while our case was an old male patient. Older age is a negative factor for DFS [2]. T and N stage, location (upper-lower lid), grade are correlated with DFS in periocular SC. In addition, large tumor size (20 mm <), diffuse pattern, multicentric origin, lobular pattern, pagetoid spread negatively affect local recurrence and distant metastasis [3].

Regional lymph node metastasis rate is 8-28% [1,4,5], the most commonly affected nodes being the preauricular, parotid, submandibular, and cervical lymph nodes.

The rate of local-regional lymph node metastasis in ocular SC is reportedly higher than that of extraocular SC [6]. Lam et al. stated that stage T2b and higher (Tumor diameter >10 mm) were related to lymph node metastasis [2], and recommended sentinel lymph node biopsy for periorbital sebaceous carcinomas >10 mm in diameter.

The recommended standard treatment is surgical excision with wide margins [7]. A minimum margin of 5 mm is considered adequate. The 5-year local recurrence rates range from 9% to 36% at 4 mm margins [8]. In case of advanced stage tumors, orbital exenteration is suggested. In addition to surgery, the roles of RT and chemotherapy (CT) have been investigated. RT was required after positive surgical margin, local-regional recurrence and/or orbital exenteration [9,10]. While studies evaluating the effectiveness of chemotherapy are limited, it can be recommended in recurrent cases [11]. In our case, no additional treatment was performed after wide local excision 7 years ago. 2 years after initial treatment, the patient presented with ipsilateral cervical lymph node recurrence. Adjuvant RT was performed with chemotherapy following regional lymph node dissection. In the literature, the rates of recurrence are 4-37% for periorbital SC [2]. The mean recurrence time is 18 months.

Conclusions

The primarily recommended treatment of the eyelid SC is wide surgical excision. Prognostic factors should be well assessed for the use of additional treatment modalities.

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