

Recurrent Pleomorphic Adenoma of Orbit: Case Report

Orbitanın Nüks Pleomorfik Adenomu: Olgu Sunumu

ABSTRACT

Pleomorphic adenoma is the most common epithelial tumors of the human lacrimal gland, comprising about 50% of all the tumors. Although malignant transformation is not frequent, intraoperative tumor spill or inadequate resection may be associated with an increase in the recurrence rate of pleomorphic adenoma. Postoperative irradiation following tumor resection appears to decrease the probability of recurrence. In this study, we present a 37-year old woman with recurrent pleomorphic adenoma of the lacrimal gland eight months after the first operation.

KEY WORDS: Pleomorphic adenoma, orbit, lacrimal gland, recurrence

ÖZ

Pleomorfik adenoma lakrimal glandın epitelyal tümörlerinin yaklaşık %50'sini oluşturan en sık görülen tümördür. Malign dönüşüm sık olmamasına rağmen, operasyonda tümörün etrafa dökülmesi veya yetersiz boşaltılması rekürrens oranlarında artma ile birlikte olabilir. Tümör boşaltılmasını takiben verilen radyasyon rekürrens oranlarını azaltabilir. Bu çalışmada, biz ilk operasyondan 8 ay sonra rekürrens gelişen pleomorfik adenomlu 37 yaşındaki kadın bir hastayı sunduk.

ANAHTAR SÖZCÜKLER: Pleomorfik adenom, orbital, lakrimal gland, rekürrens

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INTRODUCTION

Pleomorphic adenoma is one of the most frequent tumors of the lacrimal gland and constitutes 50% of tumors of epithelial origin. It can be located in the palpebral lobe or inside the lacrimal gland, because epithelial cells of the ducts and myoepithelial elements both contribute to the formation of the neoplasia (1, 4). Pleomorphic adenoma is the most common benign neoplasm of the lacrimal gland and requires a well-established clinical and therapeutic protocol to avoid the risk of malignant transformation or disease recurrence. Magnetic resonance imaging reveals a soft tissue mass with high density and shows peripheral enhancement after injection of contrast material over the superolateral portion of eye. Histologically, pleomorphic adenoma consists of myxomatous stroma and cellular areas (3, 4, 11).

CASE REPORT

A 37-year old woman was admitted to our hospital with one-month history of progressive deformation of the left eye and headache. At the physical examination, a palpable, rough, nodular mass that displaced the globe upwards and inwards and caused proptosis, 15-25 mm in diameter, was detected at the superolateral section of the left orbita. There was no pathology of the optic discs. A heterogeneous mass causing proptosis was observed on MRI. Bone destruction in the lacrimal fossa was not evident on CT. The patient was operated on at another center. The tumor in the lacrimal fossa was removed subtotally by the microsurgical approach. The histopathological findings were compatible with pleomorphic adenoma. The patient was unable to receive radiotherapy due to socioeconomic status. Eight months later, she was admitted to our hospital with a recurrent mass in the same area. The patient was evaluated with the MRI films which were obtained at the 3rd and 8th months post-operatively (Figure 1-2). An externally localized solid mass 25-30 mm in diameter, compressing the globe, located near the left lateral and superior rectus muscles was found at MRI. Histopathologic examination revealed the mass to be a recurrent pleomorphic adenoma (Figure 3). The tumor was totally removed with lateral orbitotomy at the second operation, as seen on MRI (Figure 4). We did not observe any complications at the post-operative period.

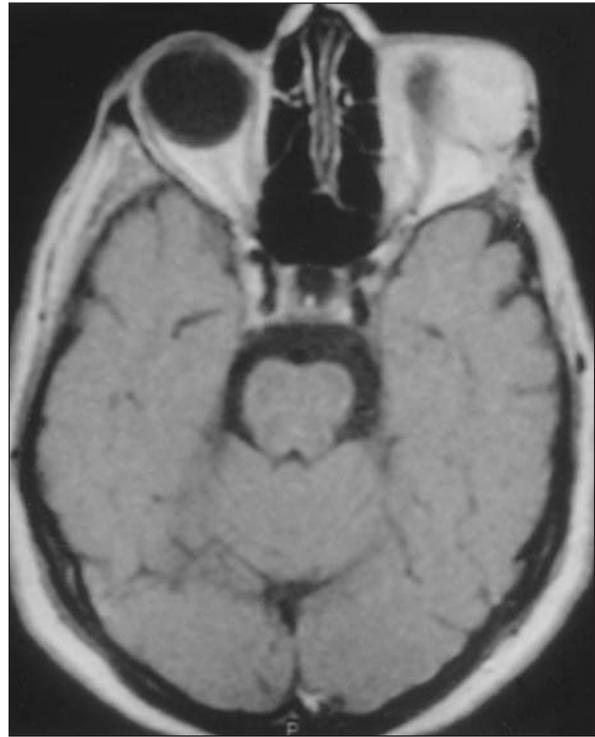


Figure 1: Contrast-enhanced T1 W image in the transverse plane at the post-operative 3rd month shows the residual enhancing mass lying laterally from the bulbus oculi. Note that the bulbus oculi is compressed by the mass.

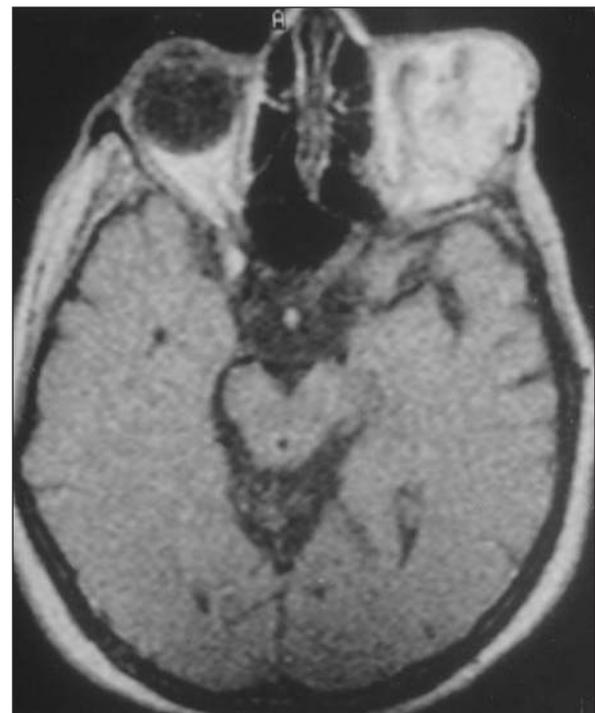


Figure 2: Contrast-enhanced T1 W transverse image at the period of the 8th month shows markedly enlarged residual mass with infiltration of surrounding structures and strong contrast enhancement.

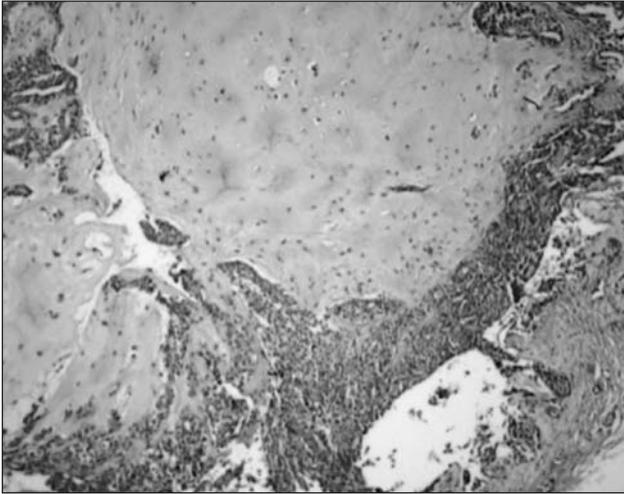


Figure 3: Pleomorphic adenoma: A mixed proliferation of both ductal epithelium, myoepithelial cells, and a hyaline/chondroid/myxomatous stroma are seen. (Haematoxylin-eosin stain, original magnification x50).



Figure 4: The mass was totally removed at the second surgical operation. Some fibrotic changes due to the surgical operation are observed as hypointense areas on the contrast-enhanced T1 W transverse image.

DISCUSSION

Neoplastic and non-neoplastic conditions may present in the lacrimal gland region. Although lacrimal gland tumors are rare and usually benign, surgical excision represents the treatment of choice

in most cases. Accurate diagnosis is crucial and the diagnostic procedure has both clinical and radiologic phases. The first consists of finding a mobile and generally non-painful neformation which slowly increases in size, occupying space in the upper external quadrant of the orbit with a possible shift of the eyeball. Other associated symptoms such as an increase in lacrimation because of the hyperactivity of the neoplastic glandular and myoepithelial cells; diplopia, which is better quantifiable by an orthoptic examination and a Hess screen; and, less frequently, an increase in intrabulbar pressure may be experienced (3, 4, 9).

CT images and especially coronal reformations are useful for the diagnosis. Radiographically visible calcifications inside the mass are rare in pleomorphic adenoma. This can help in the differential diagnosis of carcinoma, in which the incidence of such calcifications is high (1, 2).

Lacrimal gland tumors are rare; they constitute 3-10% of all lesions of the orbita. 50% of lacrimal gland tumors are epithelial. Common epithelial tumors are pleomorphic adenomas (50%), adenoid carcinomas (25%), malignant mixed tumors (7%), adenocarcinomas (4-5%), and mucoepidermoid cancers (2%). The epithelial tumors of the lacrimal gland are frequent and malignant forms are life threatening. Therefore, differentiating benign lesions from malignant ones is crucial before treatment. It has been reported that failure in total removal of the whole tumor tissue may lead to recurrence and has the risk of transformation into malignant mixed tumor (8, 9). The 5-year recurrence rate was reported to be 3% following total resection and 32% following subtotal excision (5).

Some authors have reported that radiotherapy is effective in preventing recurrences following surgery in cases with bone infiltration and results in a lower morbidity rate (7, 8). Font reported the malignant conversion rate of pleomorphic adenoma as 10% in the first twenty years following treatment and as 20% after the first 30 years (7, 10). Malignant transformation of the tumor was not evident in our patient except for the large dimensions. We thought that one of the reasons for recurrence in our patient was the lack of radiotherapy. Even when it is totally removed, a pleomorphic adenoma has the tendency to recur as in our patient and as reported elsewhere.

Although it is considered as a benign tumor, the reason of the recurrence may be related to

inoculation of the tumor cells during the operation. In order to prevent recurrence, Tang et al. have recommended tumor removal together with its capsule and no aspiration biopsies in the preoperative period (8). Wright has pointed out that tumor removal with peripheral tissues, combined with radiotherapy produced better results.

It is important to follow these steps scrupulously, because an incisional biopsy must be avoided in cases with these pathological findings. An incisional biopsy can injure the capsule, leading to dissemination of tumoral cells in the adjacent orbital tissues and exposing the patient to the risk of recurrence, and sometimes to malignant transformation of the neoplasia (1, 9).

In conclusion, in order to prevent relapse in pleomorphic adenoma surgery, the tumor should be removed with its capsule and if possible, together with peripheral tissues keeping the possibility of inoculation in mind. In addition, one should also be careful in terms of bone destruction caused by the tumor during operation.

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