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Original Article

Epithelial lacrimal gland tumors A comprehensive clinicopathologic review of 26 lesions with radiologic correlation

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Abstract

Aim: To study the prevalence, clinicopathological and radiological correlations of epithelial lacrimal gland tumors and compare these with similar published literature. The study was also designed to look at the natural history of benign mixed tumors (BMT) in regard to recurrence and malignant degeneration.
Methods: This was a retrospective study of all suspected epithelial tumors of the lacrimal gland surgically excised at King Khaled Eye Specialist Hospital (KKESH) for the period: 1983–2008. Exclusion criteria included structural lesions (dacryops) and inflammatory lesions. We included 26 cases of epithelial lacrimal gland tumors (from 24 patients). The histopathologic slides and the radiologic findings were reviewed. The corresponding demographic and clinical data were obtained by chart review using a data sheet.
Results: BMT accounted for 12/26 of the lesions while malignant lesions including adenoid cystic carcinoma (ACC) were more common (14/26). The mean age was 44.27 years (range 12–75). Commonest clinical presentation was proptosis. Median duration of symptoms in the BMT cases was 30 months and 7 months in the ACC group. The 12 BMT cases were primary in 9 and recurrent in 3 patients. The 11 ACC cases showed mostly cribriform pattern and low histopathologic grade. We had 2 cases of malignant mixed tumor (MMT) one of which arising in a recurrent tumor. One case of primary mucoepidermoid carcinoma with histopathologic grade 2 was noted. Radiologically, a well-defined appearance with bone remodeling was observed in BMT in contrast to invasive appearance with destruction in malignant lesions.
Conclusion: Our series information indicated a different distribution of benign and malignant epithelial lesions with a slightly higher rate of malignancy. BMT was the commonest benign tumor where recurrence was a squeal of incomplete surgical excision. ACC was the commonest malignant tumor with shorter duration of symptoms and radiologic evidence of invasiveness that correlated with the histopathologic features.

Keywords: Lacrimal gland, Tumor, Carcinoma, Adenoma, Adenocarcinoma

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Introduction

Lacrimal gland tumors are considered one of the challenging types of tumor which are difficult to study because of their rare incidence as they represent almost only 10% of the space occupying orbital lesions.[1] They are generally divided into 4 categories: inflammatory lesions, lymphomas, metastatic cancer, and primary epithelial tumors. The most common epithelial tumors are, in order of frequency, benign mixed tumor (BMT) or pleomorphic adenoma (PA), adenoid

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cystic carcinoma (ACC), and adenocarcinoma (ACA). Other rare variants of carcinoma are also seen, such as mucoepidermoid carcinoma. Some of the primary malignant epithelial tumors may arise from a preexisting BMT through a malignant transformation -the so called carcinoma ex pleomorphic adenoma (Ca ex PA), pleomorphic adenocarcinoma, or malignant mixed tumor (MMT) but most arise de novo. Because of rarity of these epithelial lacrimal gland tumors, most published articles describe single cases or small series, and only few large series have been reported in the English-language literature. The correlation of the histopathologic and radiologic features to the clinical presentation is important to diagnose different types of tumor and their behavior. Another important point is that lacrimal gland tumors have not been studied in detail before in our population. The aim of our study is to identify the distribution of different histologic types of primary epithelial lacrimal gland tumors in our population and to correlate the clinical, histopathologic and radiologic features of such tumors surgically treated at our tertiary referral center.

Methods

The clinical, histopathologic and radiologic data of all suspected epithelial tumors of the lacrimal gland surgically excised at King Khaled Eye Specialist Hospital (KKESH) for the period: 1983–2008 were included. A single pathologist reviewed the histopathologic slides. A single radiologist also reviewed the radiologic findings. The corresponding demographic information and clinical data were obtained by reviewing the charts of all the cases included. A specially designed Data Base Sheet was used to report all the previous information for each case. The data collected were then analyzed to calculate the prevalence of different types of epithelial lacrimal gland tumors. The correlation of the histopathologic and radiologic features to the clinical data was highlighted. Special attention was then directed toward any cases of recurrent pleomorphic adenoma to identify the pattern and occurrence of malignant change in comparison to other studies.

This study has been approved by the Human Ethics Committee-Institutional Review Board (HEC-IRB) of our institution.

Results

We have encountered a total of 28 excised lacrimal gland lesions at KKESH pathology department over a period of 26 years excluding structural (such as dacryops) and inflammatory conditions (such as Sjogren's syndrome and non-specific dacryoadenitis). 3 lymphocytic lesions were identified through tissue diagnosis accounting for about 10% only for the lesions and these were not studied further. The rest were all true epithelial lacrimal gland tumors, which were the focus of our study. 26 epithelial tumors from 24 patients (since 2 patients had recurrent lesions over their period of follow-up at our institute) have been included. The benign category consisted all of BMT accounting for 12/26 of the lesions. The overall malignant lesions including ACC were more common accounting for more than half of the total lesions 14/26. The distribution of these cases is shown in Table 1 and Graph 1.

The age ranged from 12 to 75 years with a mean of 44.27. The mean age did not differ much between the BMT and ACC groups. The lacrimal gland affected was on the right side in 14 cases and on the left side in 12. The male to female ratio was 15:11. The majority of the 24 patients were Saudis with a ratio of 16–8 non-Saudis.

The duration of symptoms ranged from 1 to 180 months with an average of 38.19 months and median of 15 months. However when this was studied in benign and malignant groups separately, the median duration in the BMT group was 30 months, while it was shorter (7 months) in the ACC group. The median duration of symptoms in the Ca ex PA was 120 months. The commonest complaints were proptosis and globe displacement in 88% of the cases each. The distribution of the presenting symptoms according to the different types of tumor is demonstrated in Graph 2. Upon examination, the commonest finding was proptosis in 21/26 cases ranging from 1 to 10 mm on the affected side (median 4 mm) followed by a palpable visible mass in 19/26.

The gross pathologic examination showed intact pseudocapsule in 10. 70% of benign lesions showed this feature. In contrast 89% (8/9) of the malignant tumors showed capsular infiltration. The only benign case with no intact capsule and an outer surface described, as ''with bosselations'' was a case of recurrent BMT. (Fig. 1a and b). The histopathologic findings of the BMT cases showed the presence of a pseudocapsule in all 12 cases (Fig. 1c), however the capsule was infiltrated by tumor extension in 5. The epithelial component was predominant (described as major or moderate component) in 5 cases only and consisted of tubular, cystic (ductal) or epithelial clusters in order of frequency (Fig. 1d). The stromal component was mostly myxomatous (6/12) followed by cartilagenous. (Fig. 1e and f). The least common type of stroma observed was osteoid and myoepithelial in 1 case each. The metaplasia of the epithelial component was mostly squamous and mainly observed in recurrent BMT.

The 12 BMT cases were primary in 9 and recurrent in 3 patients. The period until recurrence is observed was: 3 years following the initial surgery in one case where the previous histopathologic examination showed capsular infiltration indicating incomplete excision at our institute, 9 years in one of the 2 remaining cases who had his initial primary surgery elsewhere but reported as incomplete excision as well and undetermined in the third case. Follow-up of 8/12 cases was available with a mean of 90 months and no evidence of further recurrence.

Of the 11 ACC cases, cribriform pattern was noted in 6, evident by characteristic Alcian Blue staining (Fig. 2a,b), solid pattern (often with palisading) in 4 lesions (Fig. 2c,d) and tubular in 1. The histologic grade was mostly G2 (7/11) and the rest were G3. In regard to tumor behavior, vascular invasion was documented in 5/11; orbital soft tissue and/or muscle invasion was also confirmed in 5/11. This was followed by perineural invasion in 1 case only. The TNM classification

Table 1. Frequency of epithelial lacrimal gland tumors.

Histopathologic diagnosis	Number of cases	Percentage
BMT (pleomorphic adenoma)	12	46
Adenoid cystic carcinoma	11	42
Pleomorphic adenocarcinoma	2	8
Mucoepidermoid carcinoma	1	4
Total	26	100%