

# Clinicopathological survey of 23 patients with lacrimal fossa lesions in Hong Kong

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## Abstract

- **AIM:** To survey the clinicopathological correlations of lacrimal fossa lesions in the Hong Kong population.

- **METHODS:** A total of 23 patients with lacrimal fossa lesions were identified radiologically. The mean age at the time of diagnosis was 52 year-old. The medical records of patients diagnosed with lacrimal fossa lesions and subsequently underwent radiological investigations were collected and reviewed retrospectively throughout a six years period from 2000 to 2006 from three regional hospitals (Tung Wah Eastern Hospital, Pamela Youde Nethersole Eastern Hospital and Queen Mary Hospital) belonging to the Hong Kong Mega Cluster. Patient demographics, presenting signs and symptoms, imaging modalities, pathological reports and subsequent management were recorded.

- **RESULTS:** The most common initial presenting symptom was upper lid swelling (83%) followed by proptosis (13%). 19 patients (83%) had benign lacrimal fossa lesion and 4 patients (17%) had malignancy. Three of the malignancies were lymphomas and one was adenoid cystic carcinoma.

- **CONCLUSION:** Age is an important factor in considering the differential diagnosis of lacrimal fossa lesion. Lymphoproliferative diseases have the highest incidence in patients over 60 years of age while inflammatory lesions are more common in younger patients in Hong Kong.

- **KEYWORDS:** lacrimal gland; tumor; lymphoma

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## INTRODUCTION

The reported incidences and types of lacrimal fossa lesions showed great variation in previously published

literature mainly because of difference in sources of clinical data; patients recruited from primary or tertiary ophthalmic centers may have different spectrum of pathologies. They also varied from different geographic areas. Different ages of patients also affect the relative frequency of lacrimal fossa lesions. For example, malignant tumors are more common in the older adult population, comprising 63% of orbital tumors<sup>[1]</sup>. No epidemiological survey on lacrimal fossa lesions in Hong Kong has been reported up to date. The diagnosis and treatment of lacrimal fossa lesions are of great concern to ophthalmologists. Complications may arise, such as dry eyes, diplopia and squint, loss of vision, poor cosmetic outcome. Malignancy may lead to morbidity. Establishing the relationship between the pathological profiles of lacrimal fossa lesions with patients' clinical presentations and radiological findings would guide the ophthalmologists in diagnosing and managing lacrimal fossa lesions. The current study was undertaken to review cases of histopathologically or radiologically verified orbital tumors among patients who presented in eye departments at the 3 regional hospitals in the Hong Kong Mega Cluster over a six-year period. This study would provide data on the clinicopathological correlations of lacrimal fossa lesions in the Hong Kong population.

## MATERIALS AND METHODS

**Subjects** The medical records of patients diagnosed with lacrimal fossa lesions and subsequently underwent radiological investigations were identified and collected throughout 2000 to 2006 from 3 regional hospitals (Tung Wah Eastern Hospital, Pamela Youde Nethersole Eastern Hospital and Queen Mary Hospital) belonging to the Hong Kong Mega Cluster. A total of 23 patients had lacrimal fossa lesions were identified either by imaging alone or verified by histopathology if available over a six-year period in the Hong Kong Mega Cluster. There were 11 males (48%) and 12 females (52%). The age of patients diagnosed with lacrimal fossa lesions spanned from 2 year-old to 73 year-old.

**Methods** Their radiological reports were collected and intended for screening and discussion during our department's X-Ray meetings. The diagnosis was made with

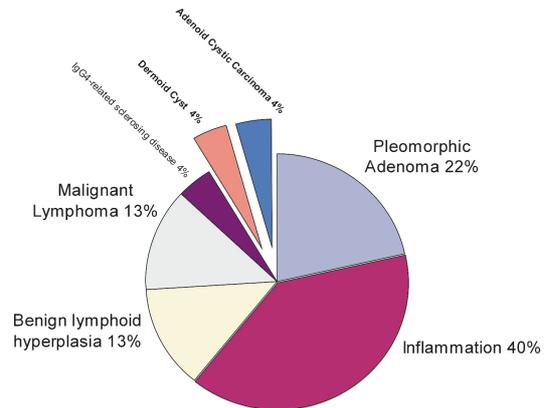
	Shields <i>et al</i> (USA) <sup>[2]</sup>	Ohtsuka <i>et al</i> (Japan) <sup>[4]</sup>	Lee S-B <i>et al</i> (Singapore) <sup>[6]</sup>	n(%) Hong Kong
Total number of patients		No. of patients		
Pathologic category	11 (10)	21 (24)	n/a	5 (22)
Pleomorphic adenoma	14 (12)	1 (1)	2 (25)	1 (4)
Adenoid cystic carcinoma	4 (4)	6 (7)	1 (13)	n/a
Pleomorphic adenocarcinoma	37 (33)	n/a	n/a	9 (39)
Dacryodentitis	7 (6)	16 (19)	n/a	3 (13)
Benign reactive lymphoid hyperplasia	16 (14)	34 (40)	2 (25)	3 (13)
Malignant lymphoma	114	84	8	23

lacrimal fossa lesion biopsy in available cases and, if not, with clinical findings correlated with imaging studies (computed tomography, magnetic resonance imaging). The patient data collected include age, gender, initial symptom and its duration, laterality (unilateral or bilateral) and the presence of any systemic cancer. The imaging method (CT or MRI) and features evaluated including appearance (ill or well defined), consistency (solid, cystic, mixed) and configuration (round, ovoid, diffuse) were recorded. If biopsies were available, samples were routinely stained with hematoxylin and eosin and examined under microscopy. Immunohistochemical techniques were used in specific tumors. The management modalities including observation, excisional biopsy, incisional biopsy, enucleation, exenteration, radiotherapy, systemic chemotherapy, local corticosteroid injection, systemic corticosteroid and any combination of these treatments were assessed. The treatment outcome for patients followed-up till November, 2006 (still under observation, recurrence or not, alive or dead with systemic cancer) were noted.

**RESULTS**

The mean age at the time of diagnosis was 52 year-old. The most common initial presenting symptom was upper lid swelling, either palpable or visualized (19 patients, 83%). The second most common symptom was proptosis (3 patients, 13%). Only 1 patient (4%) presented with incidental finding of lacrimal gland tumor in CT Scan investigating for the cause of her headache. No pain or blurring of vision was documented in patients' records as the initial presenting symptom. Amongst 23 patients diagnosed with lacrimal fossa lesion, most were benign (19 patients, 83%). There were 4 (17%) malignant tumors diagnosed by histopathology. Three of the malignancies were lymphomas and one case was adenoid cystic carcinoma. The variety of pathological profiles of lacrimal fossa lesions in our consecutive 23 patients were demonstrated in Figure 1.

Using the pathological classification previously published by Shields *et al*<sup>[2]</sup>, lacrimal gland tumors were divided into two main types, epithelial (7 patients, 30%) and non-epithelial lesions (16 patients, 70%). The most common epithelial tumor was pleomorphic adenoma (5/23 patients, 22%). Non-



**Figure 1 Pathological profiles of lacrimal fossa lesions in 23 patients**

epithelial tumors were further subclassified into inflammatory lesions (9 patients) and lymphoproliferative diseases (7 patients). Inflammatory lesions consists of pseudotumors, chronic sclerosing sialoadenitis (Sjogren's syndrome) and Mikulicz syndrome. Lymphoproliferative disease has a broad spectrum of entities which include benign reactive lymphoid hyperplasia, IgG4-related sclerosing disease and malignant lymphoma in the consecutive 23 patients. The pathological profiles of lacrimal fossa lesions were stratified according to patients' age groups. Inflammatory lesions have the highest incidence (5/11 patients, 45%) in the 40-59 years age group. Lymphoproliferative disease have the highest incidence in the 60-79 years age group (4/8 patients, 50%). The only one case of dermoid cyst belongs to a child 2 years of age. Patients' mean ages at the diagnosis of various lacrimal gland fossa pathology is summarized in Table 1.

**DISCUSSION**

There have been a number of reported large series that review orbital tumors including lacrimal fossa tumors in the general population<sup>[2-6]</sup>. Nonetheless, most reported series may have selection bias depending on the institution where the patients were recruited and different geographic locations. Lacrimal fossa lesions are uncommon and consist of a wide variety of pathologic lesions making its diagnosis and treatment challenging. The largest clinicopathologic survey is reported by Shields *et al*<sup>[2]</sup> throughout a 30-year

period. Of the 1 264 patients with orbital tumor that were studied retrospectively, 114 patients had lacrimal gland tumor. Forty-five percent were epithelial lesions and 55% were non-epithelial lesions. They revealed more malignant epithelial lesions than benign ones due to referral bias to an ophthalmic-oncology department. Similarly, malignant lymphoma is more prevalent than benign inflammatory non-epithelial lesions reported in a cohort of 86 patients throughout a 21-year period in Sapporo Medical University, Japan<sup>[4]</sup>, likely due to referral bias to their university eye centre. A comparison of the Hong Kong data from this study with other published series is summarized in Table 1. Nevertheless, we cannot disregard the fact that the incidence of malignancy in lacrimal fossa lesion rises with age. As a result of an aging population, the overall incidence of malignant lacrimal gland tumor may also be increasing. Demirci *et al*<sup>[3]</sup> studied 200 patients with orbital tumor above the age of 60 years over 20-year period and reported that 63% were malignant. Non-Hodgkin's B-cell lymphoma accounted for 16 cases (14%) in Shields et al's series of 114 patients and is the most common malignancy in lacrimal gland lesions especially in the elderly. The Japanese even reported that the incidence of malignant lymphoma has increased from 5% to 15% over a decade<sup>[5]</sup>. Such high prevalence in Japan can be attributed to the improved sensitivity of immunohistochemical and molecular techniques which help diagnosing low grade B-cell lymphoma that may have been underdiagnosed as reactive lymphoid hyperplasia previously. Moreover, a recent study indicates that the incidence of malignant lymphoma is higher in Japan comparing to Caucasian patients<sup>[7]</sup>. Infection by human T-lymphotrophic virus type-1 and even aging atomic bomb survivors are claimed to be two regional factors in Japan.

It is well known that malignant lymphomas have strong association with certain types of viral infection which differs between geographic areas. In Africa, it has been reported that the most common orbital lymphomas is Burkitt's, which is related to Epstein-Barr virus (EBV) infection.<sup>[8]</sup> In Hong Kong, EBV infection is commonly found in B cell lymphoma arising in various locations.<sup>[9]</sup> In this series of 23 patients there are 8 cases of lacrimal tumors reported in the elderly population (>60 years old) and 3 of them are diagnosed with malignant lymphoma. Therefore, malignant lymphoma is one important differential diagnosis in elderly

patients presenting with progressive upper lid swelling in Hong Kong.

Radiologically, it is challenging to differentiate a benign non-epithelial tumor from a malignant lymphoma. Inflammatory processes and lymphoproliferative disorders tend to involve the whole aspect of the lacrimal gland. They appear as diffuse enhanced lesions demonstrating soft tissue characteristics under CT and MRI<sup>[10]</sup>. Bony erosion is more likely to appear in malignancy.

This study is limited mainly by its small cohort of 23 consecutive patients over a relatively short period of six years. Since only those patients with a suspected lacrimal fossa lesion who were investigated by CT and/or MRI were identified, patients who received treatment without imaging were not included. This neglected group of patients is likely to have benign lesions such as pseudotumor with subsequent involution after a course of steroids. Nonetheless, patients have been selected from general ophthalmic clinics rather than highly sub-specialized institutes and therefore, this study may reflect more closely to the incidence of lacrimal fossa lesions in the general population of Hong Kong.

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