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Pleomorphic Adenoma of Palpebral Part of Lacrimal Gland: A Case Report

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Abstract **Case Report**

Pleomorphic adenoma is a benign salivary gland neoplasm most commonly seen in major salivary glands. It's composed of several epithelial and myoepithelial tissue elements. Pleomorphic adenoma of the lacrimal gland is a rare entity and accounts for approximately 12–25% of all lacrimal tumours. Of these 90% are seen in the orbital part and only 10% are present in the palpebral part of the lacrimal gland. We present a case report of a 33 years old female with a complaint of painless swelling in the right upper eyelid which was gradually increasing in size. A provisional diagnosis of a lacrimal gland neoplasm was made. The lacrimal mass was resected with enbloc enucleation via supratarsal fold approach. On histopathaological evaluation it showed typical morphological features of pleomorphic adenoma. After 12 months of enucleation, patient recovery was satisfactory without any signs of recurrence. This case is presented for its rarity and it highlights the need for proper diagnosis and treatment plan in the cases of benign lacrimal gland tumours.

Keywords: Pleomorphic adenoma, lacrimal Gland, Benign Salivary Gland tumours.

INTRODUCTION

Pleomorphic adenoma is a benign tumour found in lacrimal and salivary glands. BILLROTH in 1859 was the first to describe this tumour as a compound by an epithelial and a mesenchymal component. MINSENN termed it as a mixed tumour in 1874 [1]. A clinical series conducted in Wills Eye Hospital stated that tumours of the lacrimal gland constitute almost 10% of space occupying orbital lesion. 20% of them are epithelial tumours and 80% comprises either inflammatory or lymphatic lesions. Among epithelial tumours, 55% represent benign and 45% represent malignant lesions. Pleomorphic adenomas account for only 18% benign lesions [2]. This tumour mainly shows slight male preponderance with 3:2 ratios, and it is mainly seen in Third to seventh decades of life with an average age of 39 years [3]. In the literature, age of pleomorphic adenoma ranged between 6 and 80 years [4].

The mainstay treatment of this tumour is complete excision with a margin of normal tissue as incomplete resection may lead to recurrence. Incisional biopsy is contraindicated, as it can develop the risk of recurrence due to disruption of the pseudocapsule and tumour spillage [5]. This article presents a rare case report of pleomorphic adenoma of the right lacrimal gland which was surgically treated by en bloc enucleation.

CASE REPORT

A 33 year old female reported to us with complaint of painless, freely mobile hard mass present on right upper eyelid region which was gradually increasing in size and associated with increased lacrimation. On examination the patient did not have diplopia, visual acuity and globe movements were normal, mass was not fixed to skin, tarsus or periostium. Patient showed ptosis and slight proptosis. Past medical history was unremarkable. Routine blood investigations were within normal limits.

Apart from pleomorphic adenoma, a lacrimal gland duct cyst and dermoid cyst were included in the

clinical differential diagnosis. On MRI it showed enlarged right lacrimal gland mass present anterior to lateral and superolateral orbital rim measuring 2.5x1.8x2.8 cm in size. There was a slight right globe displacement in the infero-medial direction. No bony erosion and no intracranial extension were noted. Based on clinical and radiological examination a provisional diagnosis pleomorphic adenoma of palpebral lobe of lacrimal gland was made. Patient was taken under general anaesthesia after obtaining the informed consent. Supratrasal fold approach was planned. A 3cm incision was made laterally in the superior lid crease followed by sharp and blunt dissection down through orbicularis muscle (Figure 1). Enbloc enucleation of tumour along with lacrimal gland tissues was done cautiously to avoid damage to capsule (Figure 2). Primary closure of surgical site was done (Figure 3). The specimen was sent for histopathological evaluation.

Grossly, the tumour was encapsulated measuring 2.8×2.5cm. The cut surface shows solid myxoid appearance. Histopathological examination of the specimen showed a lacrimal gland with an encapsulated tumour (Figure 4). The tumour was a biphasic tumour composed of epithelial and stromal elements. The epithelial element was in the form of tubules and ducts and stroma showed abundant myxoid matrix (figure 5). Tumour showed double layered benign ducts containing intraluminal secretions and abundant myxoid stroma (figure 6).

No nuclear atypia, mitosis or necrosis was seen. Hence, a diagnosis of pleomorphic adenoma was made. The postoperative recovery was satisfactory. The patient is under a regular follow up and she is symptom free without any recurrence 1 year after surgery.

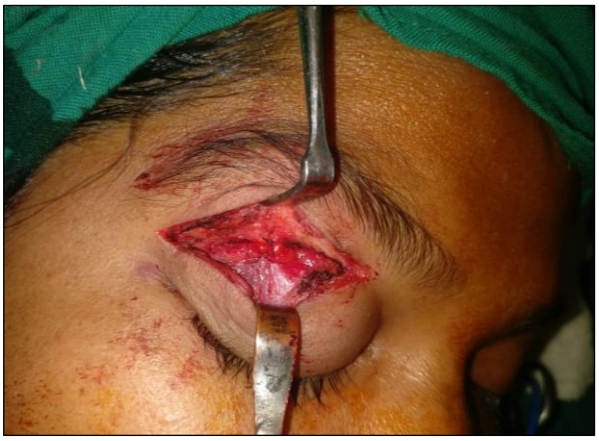

Fig-1: Supratasal fold approach and dissection

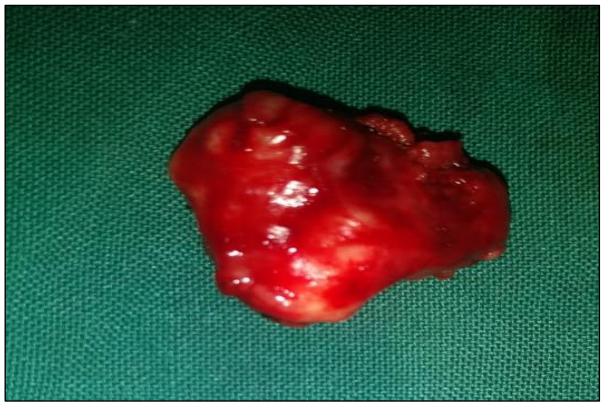

Fig-2: Enbloc enucleation of tumour along with lacrimal gland

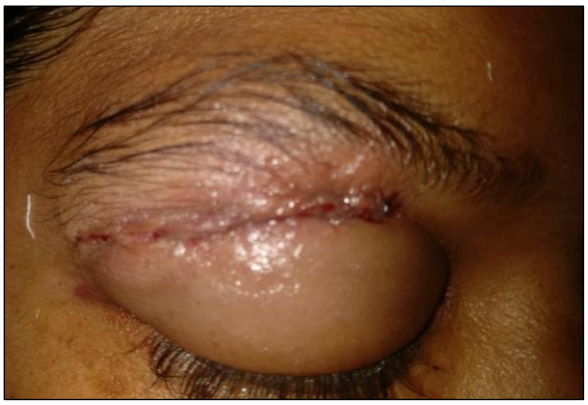

Fig-3: Primary Closure of surgical site

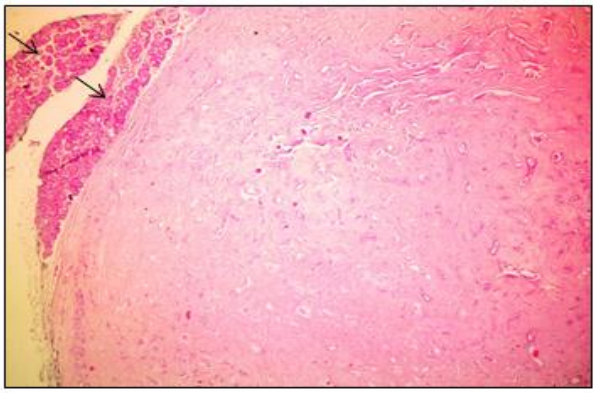

Fig-4: lacrimal gland with an encapsulated tumour (H & E x 40) Black arrow indicate lacrimal gland.

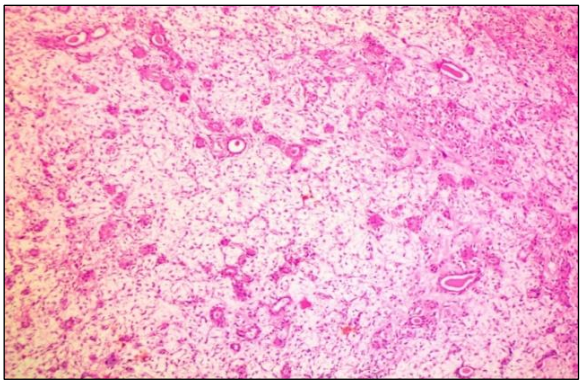

Fig-5: A biphasic tumour composed of epithelial and stromal elements (H & E x 100)

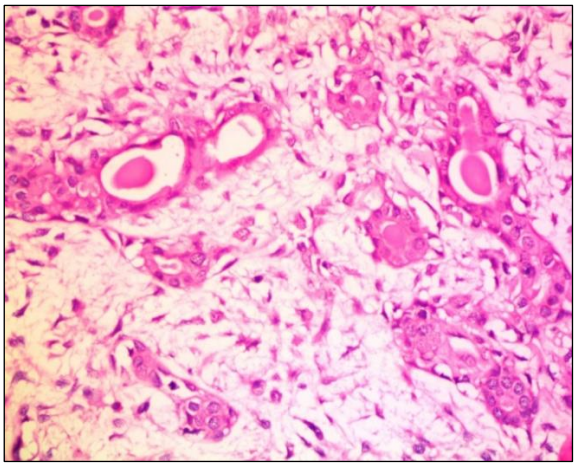

Fig-6: Tumour showed double layered benign ducts containing intraluminal secretions and abundant myxoid stroma (H & E x 400).

DISCUSSION

Lacrimal gland tumours can originate from epithelial and non epithelial components in which 30% of the lacrimal gland lesions are of epithelial origin, while pleomorphic adenoma accounts 12% of total epithelial tumours [6]. Pleomorphic adenoma of lacrimal gland can be seen in orbital lobe or palpebral lobe [7]. In our case report tumour was seen in the palpebral lobe of the lacrimal gland.

The classical clinical features associated with pleomorphic adenomas of lacrimal gland include proptosis, ptosis of eyelid, diplopia, and displacement of eyeball and disturbance of visual acuity. These symptoms have been seen over a period of more than a year [8]. In our case the patient had painless, hard slow growing mass associated with mild proptosis and ptosis without disturbance of visual acuity. Ultrasonography, skull and orbital plain films, orbital venography, CT and MRI are valuable diagnostic guides. Though for better assessment of extent of lesion, spiral CT scans are as good as MRI, but the integrity of surrounding bone and any intracranial infiltration are reported better on MRI scan [9].

Lacrimal gland tumours exhibit a lot of controversies regarding its management. ROSE AND WRIGHT [8] suggested management plan of these tumours with a most favourable rate, in which they described eight characteristics of tumour which includes duration of acute symptoms, persistent pain, sensory loss, well-defined round or oval mass, moulding of mass to the globe or along the lateral orbital wall, tumour calcification, bone invasion, and duration of symptoms related to the tumour size. According to this scoring system, if the score is equal or less than +2, it may indicate carcinoma in which incisional biopsy is acceptable. If the score is equal or higher than +3, it suggests pleomorphic adenoma and excision can be done. Based upon characteristics of our case incisional biopsy was precluded and en bloc enucleation was done.

Surgery is a mainstay modality in treatment of pleomorphic adenoma of the lacrimal gland and various approaches have been proposed [3]. Based on the case, an anterior or lateral orbitotomy could be prevailed. In our case and as lesion was present on the palpebral lobe of gland hence supratarsal fold approach was taken, as it also reduces higher chances of morbidity because not much dissection is needed if used lateral orbitotomy [10].

In our case there was a typical clinical and radiological picture depicting pleomorphic adenomas. Various authors in their literature suggested that incisional biopsy is contraindicated in such cases as it leads to chances of recurrence. Surgically removal of tumour with intact capsule and regular follow up is advisable here.

CONCLUSION

Pleomorphic adenoma of lacrimal gland is a rare disease entity that is best confirmed by clinical, radiological and histological studies. Though it is a benign tumour, but total excision of the tumour with its entire capsule is advocated. The success of treatment in such cases depends on the correct approach; complete resection and periodical follow up.

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