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Cystic pleomorphic adenoma of the lacrimal gland: a clinicopathologic study

Pleomorphic adenoma is the most common benign epithelial tumour of the lacrimal gland. It is composed of both epithelial and mesenchymal elements; the latter mainly consists of myxoid stroma. Typically, on imaging, it presents as a well-defined, solid mass in lacrimal gland fossa with bony remodeling. Internal architecture of the tumour may be heterogeneous as the myxoid stroma within the tumour can be seen as hypodense areas. Presence of cystic spaces within pleomorphic adenoma of the lacrimal gland is extremely rare and can lead to misdiagnosis. Understanding the clinical and imaging features of lacrimal gland tumour is crucial in proper management and prognostication of a case. We report the clinical, imaging, and histopathologic features of 6 cases of predominantly cystic pleomorphic adenoma of the lacrimal gland. To the best of our knowledge, there are no reports highlighting the cystic nature of pleomorphic adenoma of the lacrimal gland.

This was a retrospective study of patients with pleomorphic adenoma showing predominantly cystic spaces on imaging. The clinical details, radiographic and operative findings, and histopathology of these patients were reviewed. Cystic pleomorphic adenoma was defined where cystic component was more than solid component on imaging.

A total of 6 cases were studied. The median age of the patients was 39 years (range: 16–56 years). Four patients were males. The duration of symptoms ranged from 1 to 4 years (median: 24 months). The right eye was involved in 3 cases. Painless proptosis with inferomedial globe dystopia was present in all cases (Fig. 1A). Restriction of extraocular movement was present in 5 cases, ptosis in 2 cases, and diplopia and decreased vision in 1 case. All masses were palpable; 4 were firm, whereas 2 were soft in consistency.

Computed tomography scan in all 6 cases revealed a well-defined mass in the lacrimal gland region with bony remodeling and variable degrees of cystic spaces. An anterior solid mass with a large posterior cyst was seen in 1 case (Fig. 1B), whereas multicystic mass was seen in 5 cases (Fig. 2A, B). Surgical removal of the mass was done via lateral orbitotomy approach in 4 cases and anterior orbitotomy approach in 2 cases. In toto removal of the tumour was achieved in all cases except one, which was associated with inadvertent rupture of a cystic portion.

Microscopic examination of all specimens was consistent with pleomorphic adenoma of the lacrimal gland.

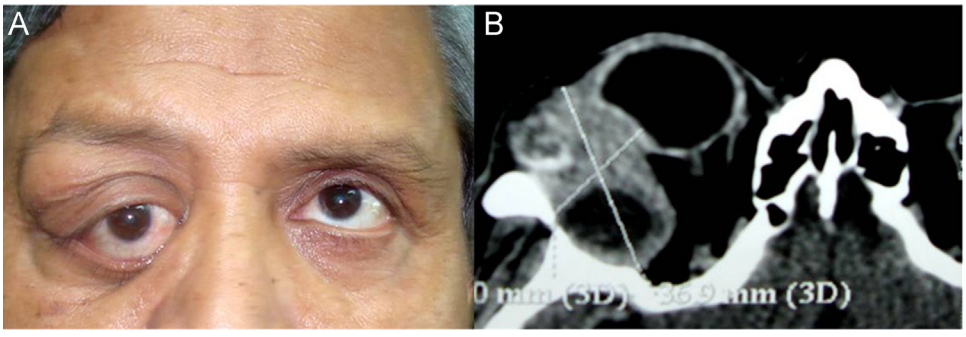

Fig. 1—(A) Clinical photograph shows mass in the right lacrimal gland region associated with inferomedial globe dystopia. (B) Computed tomography scan (axial section) shows right lacrimal gland mass with anterior solid component with calcification and posterior large cyst.

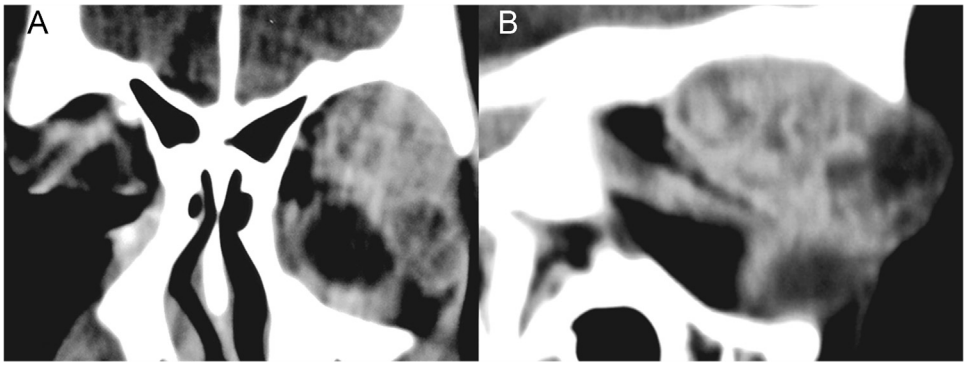

Fig. 2—(A) Computed tomography (CT) scan (coronal section) shows a large heterogeneous mass in the left lacrimal gland fossa with multiple cysts within the mass. Globe indentation is also present. (B) CT scan (sagittal section) shows a well-defined, heterogeneously enhancing multicystic mass associated with bony remodeling.

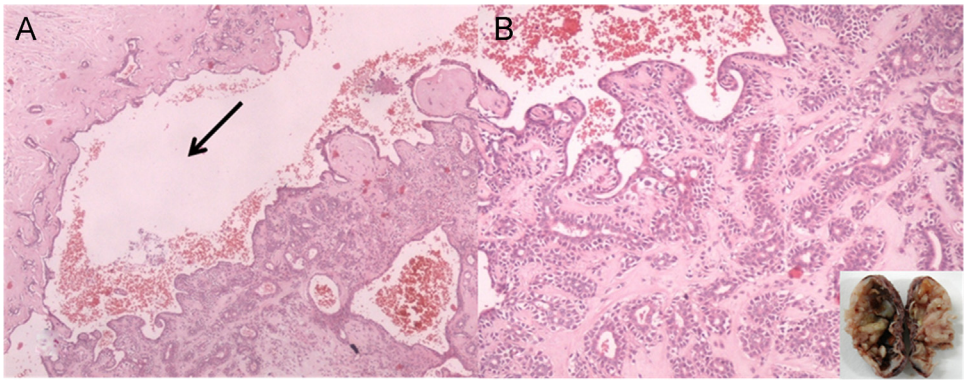

Fig. 3—(A) Photomicrograph shows a cyst (arrow) lined with squamous epithelial cells, within the stromal (left) and cellular components (right) of pleomorphic adenoma of the lacrimal gland (H&E, 100×). (B) Photomicrograph shows pleomorphic adenoma of the lacrimal gland with cellular predominance with areas of cystic degeneration (hematoxylin and eosin, 100×). Cut section showing large cystic cavity (inset).

Cystic degeneration was present in all cases (Fig. 3A). Four cases showed predominantly hyaline stroma and 2 cases showed cellular predominance (Fig. 3B). Squamous metaplasia and focal nuclear atypia was noted in 1 case. None of the patients had recurrence at 1–6 years of follow-up.

Long-standing benign tumours can undergo cystic degeneration, necrosis, or haemorrhage leading to cavities within the tumour, which changes the characteristic imaging appearance. Tumours of the lacrimal gland bear a distinct resemblance to those found in the salivary glands or other secretory glands, irrespective of their site and function. Pleomorphic adenomas with cystic changes have been reported in major and minor salivary glands.[1–3] In reported cases of cystic pleomorphic adenomas of the salivary gland, the masses were completely or predominantly cystic with small portion of solid component, unlike in our series, where multiple cysts within the solid mass were seen in most cases. In our case, the cyst was lined with a single layer of squamous epithelium (Fig. 3A). Thus, the possible origin of the cyst may be attributable to the squamous metaplasia of the tumour cells and secretions from these cells, which may lead to enlargement of the duct-like structures within the tumour. The areas of intratumoural haemorrhage or necrosis may also lead to cystic appearance on imaging.

In 2 previous reports on lacrimal gland tumours from our centre, cystic changes were found in 4% cases (2 out of 50 cases) on imaging and cystic degeneration in 36% and 16% cases on histopathology, respectively.[4,5] Preoperative differentials in cystic pleomorphic adenomas of salivary gland usually are lymphoepitheloid cyst, mucocele, and pseudocysts from a neoplastic lesions such as Warthin's tumour and adenocarcinoma. According to the incidence of lacrimal gland tumours, the possible differentials in cases with multicystic appearance are pseudocysts within a tumour-like adenoid cystic carcinoma, adenocarcinoma, carcinoma ex pleomorphic adenoma, and rarely benign lesions such as dacryops or cystadenoma.[4,6]

Cystic appearance on imaging poses a diagnostic and therapeutic challenge. In toto excision along with pseudocapsule is advised for the treatment of pleomorphic adenoma, but in such a scenario, dissection becomes difficult along the cystic wall, leading to risk of rupture and spillage of the contents. We emphasize that the diagnosis of pleomorphic adenoma should be kept in differentials of well-defined cystic masses in lacrimal fossa. Preoperative diagnosis can help in

careful and complete surgical removal of the tumour by the lateral orbitotomy approach, which provides more working space than an anterior approach and hence avoids rupture and prevents recurrence of tumour.

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Bilateral orbital lymphoma presenting as recurrence of orbital fat pad after blepharoplasty

Lymphoma is the most common orbital malignancy found in adults aged >50 years, representing approximately 10% of all orbital masses.[1–3] Diagnosis may be delayed as these tumours are often slow-growing and malleable. Low-grade, small-cell lymphomas conform to the globe and other orbital structures, thus not exerting orbital effects until proptosis or other mass effects such as diplopia, ptosis, or subcutaneous bulging ensue. Tissue biopsy is required for diagnosis, and treatment is most often local radiation.

Comparatively, orbital fat prolapse and dermatochalasis are common. The treatment of choice is elective cosmetic blepharoplasty, which is one of the most common cosmetic surgeries performed in North America.

This case study demonstrates the need for maintaining a high index of suspicion for orbital lymphoma if tissue excised during a cosmetic blepharoplasty appears abnormal.

In 2011, a 69-year-old Caucasian female was referred for slowly progressive bilateral bulging in her upper eyelids, which she stated had gradually enlarged over a 1–2-year period. She had no ophthalmological symptoms and was chiefly concerned with its cosmesis. Her medical history included osteoporosis, and her only medication was Fosavance, Merck. Her ocular history included dry eyes treated with artificial tears as needed. Her ophthalmological examination findings were normal with normal pupils, visual acuity, eye movements and lid function and no proptosis or strabismus. She was offered upper lid blepharoplasty for presumed dermatochalasis and medial fat pad prolapse, which she declined; however, she returned 1 year later wishing to pursue surgery (Fig. 1). Uneventful bilateral upper lid blepharoplasty was performed and medial fat pads were excised using Colorado needle cutting cautery. She was satisfied with her postoperative appearance (Fig. 2). The medial fat pads were noted to have a tough consistency and to be not pink or fleshy, but distinctly grey in colour. Specimens were not sent to pathology as the surgeon (R.A.) believed that the similar appearance bilaterally was indicative of a normal variant and perhaps a result of cautery-induced thermal changes. She was seen in follow-up 1 month postsurgery and discharged.

Five months after initial surgery, she returned for a small-suture cyst along her right eyelid scar. She had a moderate recurrence of the medial fat prolapse on her left and very subtle recurrence on the right. Upon palpation, this presumptive recurrent fat had a rubbery consistency. No nodes were palpated in the head and neck. The suture cyst was removed from the right scar (Fig. 3). A complete blood count and thyroid profile, including antibodies, were normal.

Bilateral orbital masses of intermediate signal intensity were seen on T1- and T2-weighted magnetic resonance sequences. The left orbital mass in the superior orbit, measuring 3.2 cm × 1.8 cm in transverse and anteroposterior dimensions, encircled the medial and superior globe

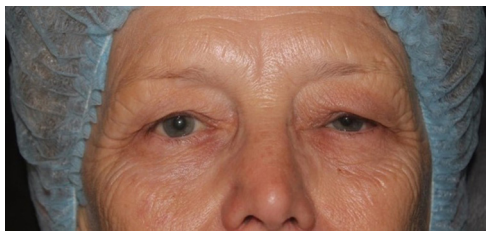

Fig. 1—Preoperative appearance before bilateral upper lid blepharoplasty.