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Case Report Volume 1 | Issue 1

KOS Journal of Public Health and Integrated Medicine
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Pleomorphic Adenoma of Lacrymal Glands: Case Reports

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Received: June 15, 2025; **Accepted:** June 19, 2025; **Published:** June 21, 2025

Citation: O Ilhami, et al. (2025) Pleomorphic Adenoma of Lacrymal Glands: Case Reports. *KOS J Pub Health Int Med.* 1(1): 1-4.

1. Abstract

Background: Pleomorphic adenoma of the lacrimal gland is a rare orbital tumor affecting young adults, with a high risk of recurrence if poorly managed. **Materials and methods:** Two cases of pleomorphic adenoma of the lacrimal gland were collected in the maxillofacial surgery department of CHU Ibn Rochd, Casablanca. **Discussion:** Pleomorphic adenomas of the lacrimal glands are very uncommon. Positive diagnosis remains difficult, and symptoms include progressive exophthalmos with or without pain, disturbance of monocular motility up to and including deterioration of vision ... Surgery is imperative, and histological study of the surgical specimen is the only way to confirm the diagnosis.

2. Keywords

Pleomorphic adenoma, Lacrimal gland, Intra-orbital tumor, Unilateral exophthalmos.

3. Introduction

Pleomorphic adenoma is a benign mixed tumor of the salivary glands, composed of epithelial and myoepithelial elements. It occurs exceptionally in the orbital cavity, developing in the lacrimal gland. In most cases, the diagnosis is made by imaging (ultrasound, CT and MRI).

This epithelial tumor has a good prognosis if surgery allows en bloc removal at a distance from the tumor capsule. Our case series including 2 cases of adenoma pleomorphic collected over a 2-year period in our department.

3. Case Reports

3.1. Case report 1

37-year-old man with no specific pathological history, presenting with ocular protrusion for 5 months with no signs of general deterioration.

Clinical examination revealed non-axial left exophthalmos with a globe deviated downwards (true dystopia) without lagophtamia, painless limitation to elevation and adduction, a significant drop in visual acuity estimated at 1/10 with an increase in ocular tone to 28 mm hg compared with 10/10 and 12mmhg respectively in the contralateral eye.

MRI imaging revealed an intra- and extra conical orbital process. The patient underwent lateral excision of the process, which was clinically dependent on the lacrimal gland.

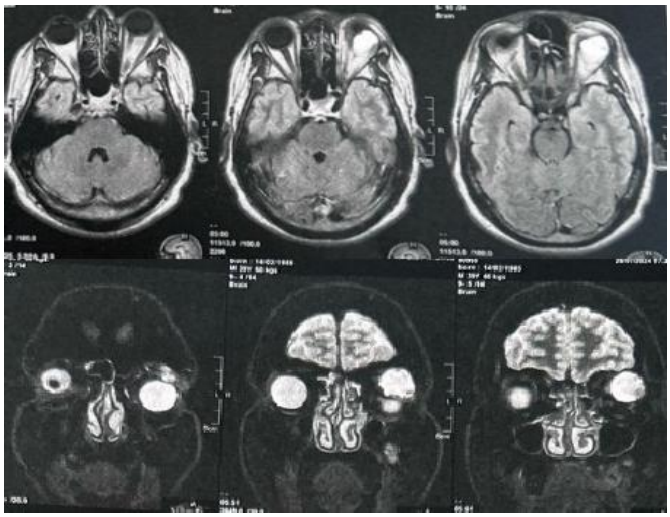
Monobloc excision of the tumor enabled histological analysis of the specimen, which concluded that it was a pleomorphic adenoma.

The clinical course was favorable (disappearance of

exophthalmos and return to normal 9/10 vision on the tenth postoperative day).

Figure 1: iconography of the patient showing exophthalmos and ocular dystopia of the left eye.

Figure 2: MRI (axial and coronal slices) showing the intra- and extra conical process in the left orbit, responsible for exophthalmos and pushing the eyeball downwards.



3.2. Case report 2

A 35-year-old man presented with left exophthalmos of progressive onset over the past 18 months, accompanied by decreased visual acuity.

The patient had already undergone surgery for the same symptomatology at the age of 25 (i.e. before 10 years). The final anatomopathological examination of the exeresis specimen revealed a pleomorphic adenoma of the lacrimal gland measuring 3cm.

Clinical examination revealed non-axial left exophthalmos with downward ocular dystopia, with no limitation of oculomotor movement or diplopia, and a significant drop in visual acuity to 5/10 et un larmoiement . Palpation revealed a firm, painless mass on the supraorbital rim responsible for false ptosis.

MRI imaging showed a left superior-external extra conical mass responsible for grade 1 exophthalmos, with adjacent bone involvement and nodular soft-tissue formations of similar characteristics, suggesting recurrence or malignant degeneration of the adenoma. Excision by left lateral orbitotomy and creation of a bone flap (taking up the old scar) revealed a friable mass at the level of the external border. Histological analysis of the specimen concluded a pleomorphic adenoma.

Clinical outcome was favorable. A follow-up CT scan at one year showed no signs of recurrence.

Figure 3: iconography of the patient showing the left exophthalmos, left ptosis and inferomedian shift of the left eyeball due to a left lacrimal gland tumor.

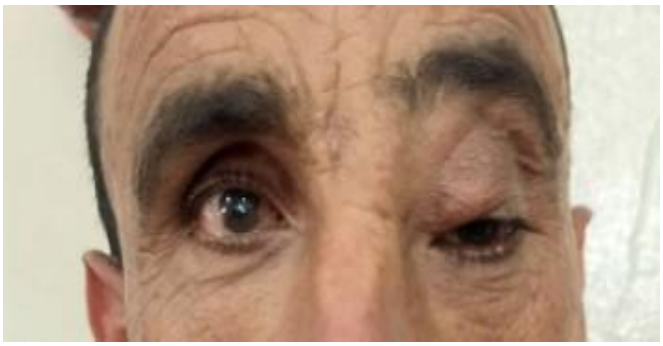
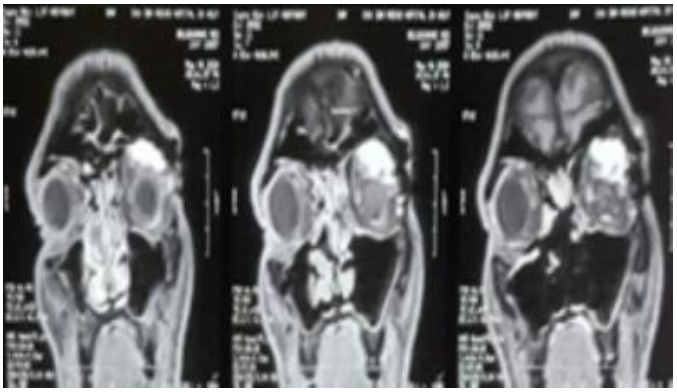


Figure 4: MRI imaging showing a left superior-external multilobulated extra conical mass of tissue density in hypersignal after gadolinium injection, measuring 30*26 mm and extending over 18 mm, associated with bone involvement and nodular formations in the soft tissues opposite the mass of similar characteristics.



4. Discussion

Pleomorphic adenomas are benign tumours frequently found in the parotid gland (80% of cases).

Rarely (3 to 5% of cases), the pleomorphic adenoma may be located in the intraorbital region or, more precisely, in the lacrimal gland.

The average age is between 35 and 50. Invasion is always unilateral, most often on the right. Its insidious appearance explains the delay in diagnosis

4.1. Clinical presentation

The clinic differs according to the part of the lacrimal gland affecte:

A. Orbital part: painless, unilateral, non-pulsatile, irreductible exophthalmos proportional to the volume of the tumour.

The ocular protrusion is non-axial however the eyeball is pushed inwards and downwards, sometimes causing vertical diplopia, and often partial ptosis.

B. palpebram part (rare): firm mass without ocular protrusion, and the lesion is apparent under the conjunctiva after simple eversion of the upper eyelid.

Some forms have been described, notably after exposure to irradiation for a malignant tumor such as retinoblastoma. As the tumour progresses, a mobile mass may be palpated under the skin, with or without local oedema.

In addition, other ophthalmological signs may be present: reduced visual acuity due to astigmatism or acquired hyperopia, lagophthalmos due to keratitis, visual field impairment due to compression of the posterior pole, scotomas or fundus abnormalities.

Rarely, this tumor may have a painful character. However, this sign appears much more frequently in malignant tumors or in cases of dysplasia.

Other atypical forms including a painful subcutaneous nodule, calcification and bone erosion have been reported in the literature.

4.2. Radiological presentation

In such cases, the imaging of choice for diagnostic affirmation is orbital CT with injection CT is considered superior to MRI for analysis of changes in the bone wall.

CT scan of the orbit shows diffuse enlargement of the lacrimal fossa with cortical integrity and a broadly rounded, well- circumscribed lesion, isodense in relation to the extraocular muscles, with moderate and homogeneous contrast, located in the superior-external orbital angle, i.e. in the region of the lacrimal gland, and extending rather posteriorly, with downward and inward displacement of the globe

The surface of the eyeball adjacent to the tumor may become irregular in the course of its natural evolution.

Bone lysis, sometimes leading to a true defect, may be observed, particularly in cases where the tumor extends to the roof or lateral wall.

4.3. Pathological diagnosis

Macroscopically, the tumor is circumscribed, usually over 2 cm in size, well delimited (pseudocapsule) and globular. Occasionally, extracapsular tumor islands may be observed. intracapsular tissue is friable.

Diagnostic confirmation is obtained by anatomopathological examination of the surgical specimen. The microscopic appearance is similar to that of other localizations, with two components: glandular, with cords and tubes or other cavities filled with a mucoid substance, and conjunctival, consisting of connective tissue of variable density. The scarcity of mitoses and the absence of necrosis avoid any confusion with a malignant process.

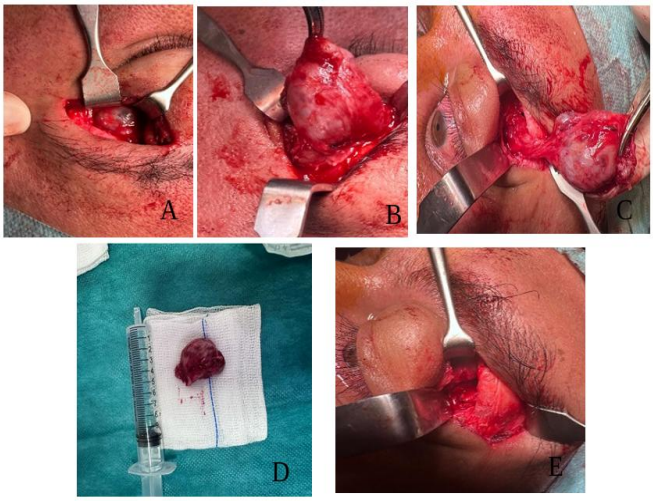
4.4. Treatment and evolution

Treatment is based on surgery using a lateral orbitotomy or transconjunctival approach. The surgical approach must be sufficiently wide to allow monobloc excision of the tumour and its capsule, which is mainly responsible for recurrence or even malignant degeneration.

A ring of healthy glandular tissue surrounding the tumour should be removed, while respecting the palpebral lobe to avoid postoperative keratoconjunctivitis; similarly, any nearby periorbititis should be removed.

The lateral approach is most often used, but a wider superolateral approach with orbitotomy is sometimes necessary for large tumors, enabling exeresis that must be complete and en bloc.

Figure 5: illustrations of the patient's surgical procedure (case 1) showing in A-B and C the lateral approach to the orbit, the mass abutting the external wall, and the various sequences of its removal. D: showing the well-rounded tumor with its capsule, E showing the cavity after removal of the mass and the absence of bone lysis.



Follow-up is essential to detect recurrence. These recurrences (19-32% of cases), sometimes possible after 20 years or more after the initial surgery, are the result of fragmented biopsies, classically contraindicated.

5. Conclusion

Pleomorphic adenomas are rare benign tumors requiring total surgical excision, including the capsule, and a surgical approach allowing en bloc removal. Clinical and radiological monitoring for several years should be systematic.

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