RESEARCH PAPER

Intact Removal Of Huge Lacrimal Gland Tumor With Minimal Invasive Lateral Orbitotomy

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ABSTRACT

Objectives: Lacrimal gland tumors are uncommon and make up about 10% of all orbital masses. Among these, the most frequent benign type is pleomorphic adenoma, which comprises nearly half of all epithelial tumors in that area. The standard approach involves surgically excising the entire pleomorphic adenoma, including its surrounding pseudocapsule and a rim of adjacent orbital tissue. We outline the diagnostic and management process done to patient, resulting satisfactory result.

Methods: A 28-year-old woman presented with a progressively protruding of her right eye, causing blurred vision, globe displacement, and ocular motility obstruction. Contrast CT Scan imaging show regular border mass arising from the lacrimal gland without molding, bone involvement, nor calcification. Complete excision using a minimal invasive lateral orbitotomy approach was performed. Histopathology examination confirmed of pleomorphic adenoma without sign of malignancy. Follow up two months after surgery showed no right eye protrusion with improvement of visual and ocular motility.

Results: Management of lacrimal gland tumors depends on the initial appearance, complaints, clinical condition, and imaging results. Treatment of benign cases (pleomorphic adenoma) with complete excision of the tumor, results in clinical improvement and a good prognosis with low recurrencies.

Conclusion: This case highlights the diagnostic and management of lacrimal gland tumor (pleopmorphic adenoma). Initial diagnosis and subsequent action plans can influence patient outcomes and prognosis.

Keywords: lacrimal gland tumor**,** pleomorphic adenoma, orbitotomy

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INTRODUCTION

The primary lacrimal gland rests in a shallow depression of the orbital region of the frontal bone, separated from the main orbital cavity by a layer of fibroadipose tissue. It is anatomically divided into two sections, known as the orbital and palpebral lobes. This gland produces the aqueous part of the tear film, which is rich in enzymes and proteins like lysozyme, lactoferrin, and immunoglobulins. Its blood supply is provided by the lacrimal artery, a branch of the ophthalmic artery, and its function is regulated by the lacrimal nerve, a branch of the ophthalmic division of the trigeminal nerve.[1] The lacrimal gland is a small, almond-shaped, two-lobed gland roughly 2 cm in length, located in the lacrimal fossa of the superolateral orbit. It functions as an eccrine gland by producing a watery fluid that lubricates and nourishes the eye, and it contains lysozyme, an enzyme that helps kill bacteria. Various types of tumors, including metastases, can develop in the lacrimal gland, representing between 5% and 25% of all orbital tumors. Most primary tumor in this gland emerge from the orbital lobe, where the gland is attached to the orbital rim near the lacrimal fossa. The most frequently encountered benign tumor is the pleomorphic adenoma—a benign mixed tumor composed of both epithelial and mesenchymal tissue.[2]

A study reported in Denmark, lacrimal gland tumors are exceptionally uncommon—only about one case per million people is reported annually. They account for

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roughly 10% of all orbital masses, and most of these growths are benign. Patients typically experience facial asymmetry caused by displacement of the eyeball, along with drooping eyelids, reduced eye movement, and swelling of the lacrimal gland. [3] A review of the literature found that about one-tenth of all orbital masses are located in the lacrimal gland. When looking at solid masses within this gland, roughly one-fifth arise from epithelial cells while the vast majority come from non-epithelial tissues. Of the epithelial tumors reported, a bit more than half were benign and the remainder were malignant. Among these malignant cases, most are adenoid cystic carcinomas, followed by pleomorphic adenocarcinomas; a smaller portion are primary adenocarcinomas that develop on their own, with an even tinier fraction being mucoepidermoid carcinomas or other miscellaneous malignant epithelial types.[4]

Among the epithelial tumors of the lacrimal gland, pleomorphic adenoma—often referred to as a benign mixed tumor. It tends to appear slightly more often in men than in women and is usually seen in individuals during their 40s or 50s. This tumor is only surrounded by a pseudocapsule and grows slowly by expanding, a process that can gradually create a cavity in the bone of the lacrimal fossa. As it enlarges, the tumor prompts the periosteum to lay down a thin layer of new bone, a phenomenon known as cortication, though the nearby orbital bone remains unaffected. Typically, the condition is painless. However, if a pleomorphic adenoma persists for a long time or is not completely removed, it may undergo malignant transformation into adenocarcinoma—termed carcinoma ex pleomorphic adenoma—which then tends to grow rapidly following a period of relative inactivity. Additionally, when these adenomas recur in the orbit, they may also give rise to malignant tumors such as adenoid cystic carcinoma.[5]

This case report presents an adult with relatively young age-patient with pleomorphic adenoma confirmed by histopathological finding, emphasizing the diagnostic process and treatment considerations, resulting in delightful outcome.

CASE ILLUSTRATION

A 28-year-old woman presented to the ophthalmic oncology clinic with a two-year history of noticeable protrusion of her right eye, accompanied by blurred vision in that eye for the past year. She reported no pain in the periorbital or head regions and no other systemic complaints, with her left eye remaining unaffected. The patient's medical history was unremarkable, with no previous treatments, eyewear usage, or prior ocular issues. On physical examination, her vital signs were within normal limits. Visual acuity in the right eye was reduced to counting fingers at two meters, while the left eye maintained a normal 5/5 vision. Colour vision deficiency was found with Ishihara plates in the right eye, with normal results in the left eye. Intraocular pressures in both eyes were normal. Inspection revealed that the right eye was noticeably protruding, with the globe displaced inferomedially and a firm-painless-well defined palpable mass at the supero-temporal orbital rim. There was no hypoesthesia on right/left side of patients face, this examination was done to rule out trigeminal (V1-V2) nerve involvement. Extra ocular movements in the right eye were restricted in all directions, in contrast to normal movements in the left eye. Hertel exophthalmometry measurements (26-112-18) indicated significant proptosis of the right eye. (Figure 2.1)

No lagophthalmos was observed in the right eye; however, a relative afferent pupillary defect (RAPD) was present, while the anterior segment examination of both eyes was otherwise unremarkable. During the posterior segment examination, the margin of the right optic disc appeared indistinct, although all other details were within normal limits, while the posterior segment of the left eye was normal.

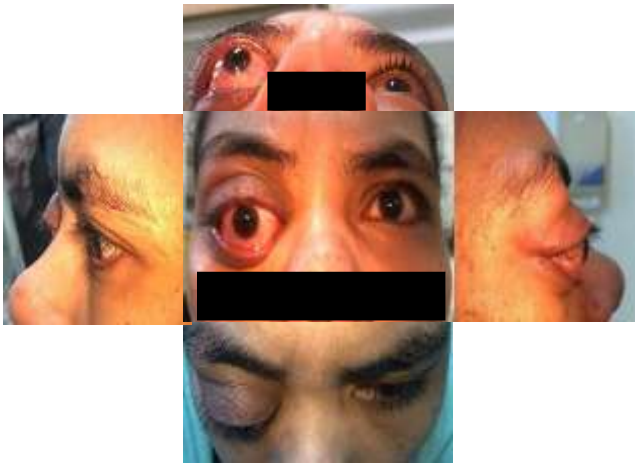

Figure 2.1 Axial proptosis with inferomedial globe displacement. A fix and firm mass on supero-temporal of patient right eye orbit. (Picture was taken with patient's consent, Courtesy of POSA Mata RSUD Dr. Soetomo, 2025)

Contrast orbital CT Scan imaging was done to support the diagnostic process, A solid homogenous round mass with well-defined margins, measuring approximately 3.3 x 3.3 x 3.0 cm, is observed in the extraconal space of the right orbit in the superolateral region, with features suggestive of an origin from the lacrimal gland and demonstrating contrast enhancement, without moulding of the mass, calcification, nor invasion of bone. The mass is contiguous with the supero-posterior aspect of the right globe, exhibiting clear borders; it displacing the globe antero-inferiorly (Figure 2.2)

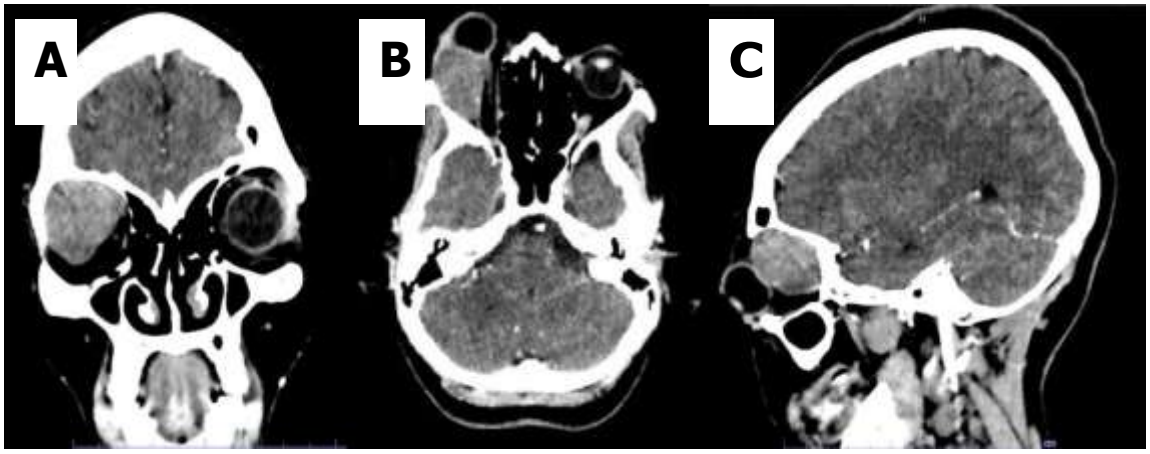

Figure 2.2 Contrast Orbital CT-Scan. (A) Axial, (B) Coronal, (C) Sagital, respectively shown well defined gland lacrimal gland tumor with mild contrast enhancement (Courtesy of RSUD Dr. Soetomo, 2025)

Based on the evaluation of the patient's complaints, clinical condition, and imaging results, the lacrimal gland tumor in the right eye was determined to be a benign lesion, most consistent with a pleomorphic adenoma. A total tumor excision via lateral orbitotomy was planned, with the patient prepared for the procedure under general anesthesia.

During the surgery, injection of local anesthesia at the marked area, and then a Stallard-Wright incision was made. cryoprobe assisted dissection was carried out to identify and separate the mass from the surrounding tissue, then the mass was flipped out of the orbit. The mass was excised intact without compromising the capsule and with minimal manipulation of the adjacent structures. The excised mass measured approximately 3 x 3 x 4 cm with an intact capsule and was sent for histopathological examination. The orbital cavity was subsequently irrigated with povidone iodine and sterile water, and the septum and orbicularis were sutured, followed by skin closure. Finally, a compression dressing was applied to the right eye. (Figure 2.3)

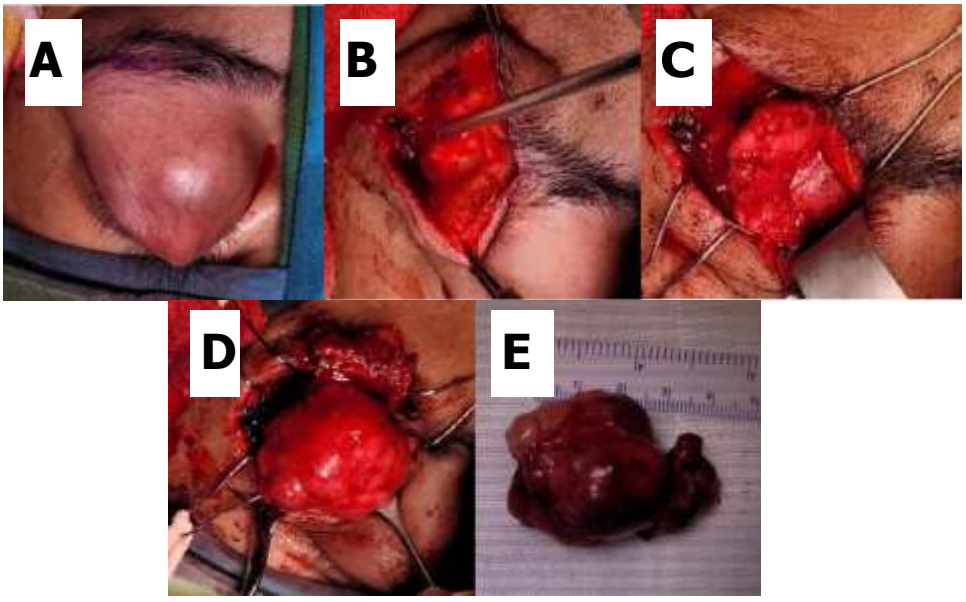

Figure 2.3 Technique of lacrimal gland tumor removal. (A) Sub brow marking and Stallard-Wright incision was made. (B),(C),(D) Cryoprobe assisted dissection was carried out to identify and separate the mass from the surrounding tissue. (E) The mass was excised intact without compromising the capsule and with minimal manipulation of the adjacent structures. (Courtesy of RSUD Dr. Soetomo, 2025)

Following surgery, the patient received antibiotic and steroid injections until the third postoperative day, during which the surgical wound, visual acuity, and pupillary

response were assessed. On that day, the right eye's visual acuity was constant at 2 meters counting finger, accompanied by a mildly dilated pupil with a diminished light reflex and a drooping upper eyelid. Consequently, the patient was discharged with prescriptions for both oral and topical antibiotics as well as oral steroids. From the latest outpatient clinic appointment, patient visual acuity was improved to 5/40, color vision improvement, right eye ptosis and ocular motility improvement (Figure 2.4)

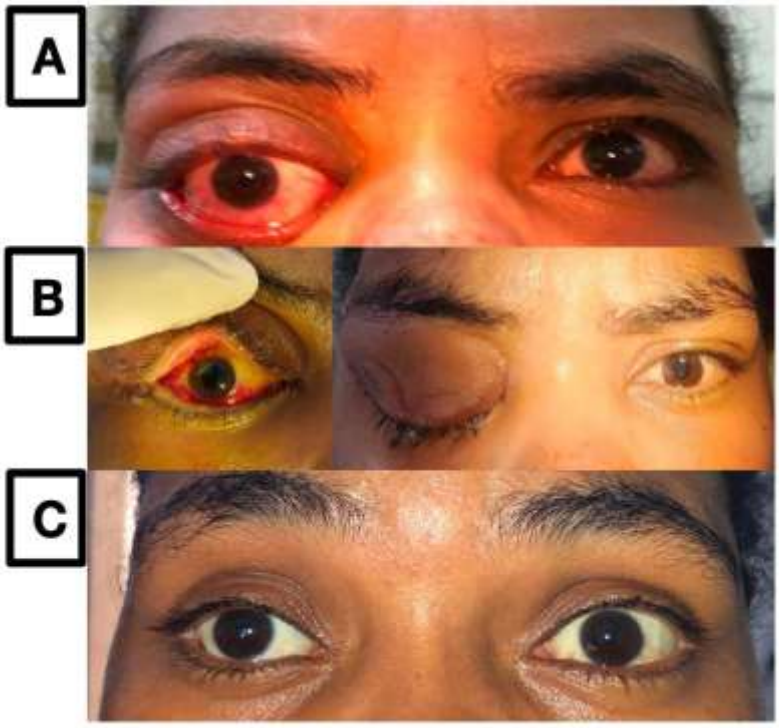

Figure 2.4 Patient pre operative condition and post operative outcome. (A) Pre operative condition, (B) Post operative condition, (C) Latest evaluation (Picture was taken with Patient's consent. Courtesy of RSUD Dr. Soetomo, 2025)

The completely excised tissue was subjected to detailed histopathological examination following routine fixation, processing, and hematoxylin–eosin staining. Microscopic evaluation revealed a well-circumscribed and well-demarcated tumor, partially encapsulated and separated from the surrounding normal salivary gland parenchyma by a thin fibrous capsule. The lesion was composed predominantly of duct-like tubules and glandular structures, some cystically dilated. These tubular formations were lined by an inner layer of cuboidal to columnar epithelial cells and an outer layer of myoepithelial cells, confirming the biphasic cellular architecture typical of salivary gland neoplasms. The luminal spaces of several ducts contained eosinophilic material suggestive of secretory activity.

The intervening stroma displayed abundant myxoid and focal chondromyxoid matrix, interspersed with spindle-shaped and stellate cells within a loose extracellular background. Areas of squamous metaplasia were noted within certain ductal components, without evidence of keratin pearl formation or dysplasia. There were no features of cytologic atypia, increased mitotic activity, necrosis, capsular invasion, or perineural infiltration. Based on these histomorphological findings, the lesion was diagnosed as pleomorphic adenoma (Figure 2.5).

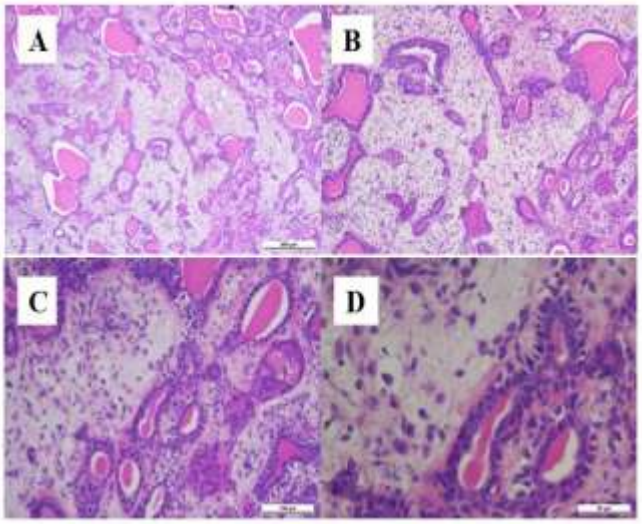

Figure 2.5 Excised tissue histopathological finding. (A) 40x magnification, showing cystically dilated tubule. (B) 100x magnification, showing squamous metaplasia. (C) 200x magnification, showing stroma characterized by myxoid matrix with stellate cells. (D) 400x magnification, the tubules were lined by epithelial and myoepithelial cells. (Picture was taken with Patient's consent. Courtesy of RSUD Dr. Soetomo, 2025)

DISCUSSION

A 28-year-old woman presented to the ophthalmic oncology clinic with a two-year history of noticeable protrusion of her right eye, accompanied by blurred vision in that eye for the past year. Visual acuity in the right eye was reduced to counting fingers at two meters, colour vision deficiency was found with Ishihara plates in the right eye. Inspection revealed that the right eye was noticeably protruding, with the globe displaced inferomedially and a firm-painless-well defined palpable mass at the supero-temporal orbital rim. There was no hypoesthesia on right/left side of patients face. Extra ocular movements in the right eye were restricted in all directions, in contrast to normal movements in the left eye. Hertel exophthalmometry measurements (26-112-18) indicated significant proptosis of the right eye. A relative afferent pupillary defect (RAPD) was present on right eye, while the anterior segment examination of both eyes was otherwise unremarkable. During the posterior segment examination, the margin of the right optic disc appeared indistinct, although all other details were within normal limits.

The majority of lacrimal gland masses represent inflammation, known as dacryoadenitis, which presents with acute inflammatory signs and typically responds well to anti-inflammatory treatment. In cases where lacrimal gland enlargement occurs without these inflammatory features, the underlying cause is most often tumor such as lymphoproliferative disorder, as many as 50% of orbital lymphomas originate in the lacrimal fossa, while only a small fraction of lesions in that area are epithelial tumors.[6] Lacrimal gland tumors are typically divided into two main categories: epithelial and non-epithelial. Non-epithelial lesions are more frequently encountered, comprising between 70% and 80% of solid lacrimal gland masses, with lymphomas accounting for nearly 37% of all malignant tumors in this region. In contrast, epithelial tumors exhibit a more balanced distribution, with roughly 55% being benign and 45% malignant, although this proportion can be even more skewed toward benign lesions in certain Asian populations, where benign epithelial tumors may represent up to 72-76% of cases. Within the benign group, pleomorphic adenoma—also known as a benign mixed tumor—is the most common lesion, while among malignant epithelial tumors, adenoid cystic carcinoma is the predominant type, making up approximately 60% of cases; the remainder includes pleomorphic adenocarcinomas, primary adenocarcinomas, mucoepidermoid carcinomas, and various other less common malignant entities.[7]

On orbita CT scans, adenoid cystic carcinomas tend to exhibit a higher frequency of bone invasion, tumor calcification, and the wedge sign, whereas pleomorphic adenomas are more commonly associated with bone remodeling. Additionally, on T2-weighted MRI, pleomorphic adenomas are more likely to display clear, well-defined margins, a lobulated appearance, heterogeneous contrast enhancement, and increased signal intensity. [8] Whereas in our case, A solid heterogenous round mass with well-defined margins, measuring approximately 3.3 x 3.3 x 3.0 cm, is observed in the extraconal space of the right orbit in the superolateral region, with features suggestive of an origin from the lacrimal gland and demonstrating contrast enhancement, without moulding of the mass, calcification, nor invasion of bone. The mass is contiguous with the supero-posterior aspect of the right globe, exhibiting clear borders; it displacing the globe antero-inferiorly.

According to the Rose and Wright criteria, lacrimal gland masses are evaluated using both clinical features and radiological findings. Clinically, the assessment considers whether the lesion has developed acutely in less than 10 months, whether there is persistent pain, and if there is any reduction in sensory function. Radiologically, the focus is on detecting an ill-defined round or oval mass that molds to the globe or lateral orbital wall, along with signs such as tumor calcification, bone invasion, and a correlation between the tumor size and symptom duration—where a larger mass is associated with a shorter history of symptoms. Each of these parameters is assigned a value of -1 if present and +1 if absent. A cumulative score ranging from +3 to +8 suggests that the tumor is benign, whereas a score between -8 and +2 indicates a malignant process. The Rose and Wright system demonstrates a sensitivity of 64%, a specificity of 93%, and an overall accuracy of 86%. [9,10] The diagnosis of a pleomorphic adenoma in the lacrimal gland is typically established through clinical examination and imaging studies. When these findings clearly indicate the diagnosis, performing an incisional biopsy is generally avoided to prevent disruption of the tumor's capsule.[11]

As in our case, patient was presented with chronic growing lesion >2years (+1), no pain (+1), no sensory function defect (+1), On CT Scan Imaging were found well defined round mass (+1), no molding to the lateral orbital wall or globe (+1), no sign of calcification (+1), no bone invasion (+1), and relatively large tumor with long symptoms (+1). The patients Rose and Wrights algorithm score was +8, strongly indicated of benign lesion (pleomorphic adenoma).

The recommended treatment involves complete excision of the pleomorphic adenoma along with its pseudocapsule and a surrounding rim of orbital tissue. It is advised not to perform a preliminary biopsy, as early studies indicated that incising the capsule for biopsy led to a 32% recurrence rate. Moreover, when recurrences occur, there is an estimated 10% risk per decade of the tumor undergoing malignant transformation into carcinoma ex pleomorphic adenoma. [6] Other reports stated 20% of breached -capsule pleomorphic adenoma undergone malignant degeneration and recurrence rate of 20% - 45%. [12,13] Thus far, the most effective treatment remains the prevention of pleomorphic adenoma recurrence by performing a complete excision while preserving the integrity of its pseudocapsule. When comprehensive

imaging is available, a biopsy is unnecessary. Once a pleomorphic adenoma recurs, it becomes considerably more challenging to manage, often necessitating multiple surgical procedures over a broad anatomical region and, in some cases, may eventually require radical intervention such as orbital exenteration. [14] Intact excision could minimize recurrence rate to 3%. [15] A study stated that carcinoma ex pleomorphic adenoma with one in four patients experienced tumor-related death at an average of 2.8 years post-diagnosis, with an overall survival rate of 63% observed at both the 5- and 10-year marks. The prognosis for carcinoma ex pleomorphic adenoma was considerably poorer, especially in cases where there was a history of incomplete excision of a pleomorphic adenoma, the presence of a highly invasive malignant component, and signs of perineural invasion.[16]

A lateral orbitotomy is performed to reach lacrimal lesions that necessitate complete removal of the lacrimal gland, such as pleomorphic adenoma, and to access lesions situated in the middle and posterior regions of the orbit. [17] Traditionally, surgeons remove well-circumscribed orbital lobe masses using a lateral wall osteotomy. This approach provides easier access to the gland's posterior pole, where the lacrimal neurovascular bundle enters, allowing for clear visualization of the blood supply and effective control of bleeding before the tumor is mobilized and excised intact along the junction between the orbital and palpebral lobes. However, removing the mass without performing a lateral osteotomy offers benefits such as shorter surgery time, lower complication rates, and faster recovery. Additionally, leaving the bone intact helps prevent intraosseous seeding in case the lesion ruptures during removal, as the unaltered cortical bone acts as a natural barrier to tumor spread. It would appear that almost all well-defined masses within the orbital lobe of the lacrimal gland can be safely excised without the need for lateral osteotomy, with a concomitant reduction in postoperative morbidity and faster recovery.[18]

In general, tumors of the lacrimal gland closely resemble those of the salivary glands, reflecting their common origin during embryonic development. [19] Histopathologic evaluation of pleomorphic adenomas reveals a proliferation of both epithelial and myoepithelial elements that organize into spindle-shaped as well as glandular or duct-like formations containing lumens. These tumor components are typically encased by a pseudocapsule of normal tissue, although there can be small areas where tumor cells have microscopically breached this boundary.[20] Lacrimal gland pleomorphic adenomas exhibit a pathological mixture of glandular epithelial cells, myoepithelial cells, and interstitial components, which may be attributed to an imbalance between cell death and cell growth leading to excessive proliferation.[21]

Pleomorphic adenoma, commonly referred to as a benign mixed tumor, is the most frequently encountered epithelial neoplasm in the lacrimal gland. It appears somewhat more often in men than in women and generally emerges in individuals during their fourth or fifth decade. Patients typically exhibit a slowly progressive, painless downward and inward displacement of the eyeball, accompanied by axial proptosis. These signs generally persist for over a year. On examination, a firm, lobulated mass can often be felt near the superolateral orbital rim, while orbital imaging commonly reveals an enlarged or expanded lacrimal gland fossa. The lesion itself usually appears well defined on imaging, although it may display a subtly nodular contour. The tumor is characterized by a pseudocapsule and exhibits slow, expansile growth. As it gradually enlarges, it may carve out a cavity in the bone of the lacrimal fossa, prompting the periosteum to deposit a thin layer of new bone—a process known as cortication—while leaving the adjacent orbital bone intact. Typically, this condition is not accompanied by pain. [5,6] Most of the lacrimal gland pleomorphic adenoma arise from the orbital lobe, the occurrence of palpebral pleomorphic adenoma is relatively rare; a retrospective study documented 43 cases over a period of 41 years of medical records.[22]

As mentioned above, patients clincical presentation and examination suggested of a benign lacrimal lesion, rose and wright algorithm were used and the result were highly suggested of pleomorphic adenoma. Total excision of the tumor was done without prior biopsy. Lateral orbitotomy without osteotomy was performed to excised tumor with the capsule intact, lateral orbitotomy without osteotomy enchance the patient recovery process. Histopathological evaluation was in accordance with rose and wright algorithm, showing of pleomorphic adenoma.

CONCLUSION

In conclusion, this case highlights that complete surgical excision of a pleomorphic adenoma of the lacrimal gland can lead to a favorable outcome. The patient experienced a marked improvement compared to previous pre operative condition. The preservation of the tumor's pseudocapsule contributing to a low risk of recurrence. This successful management underscores the importance of early and precise diagnosis, followed by meticulous surgical intervention, in restoring function and enhancing quality of life for patients with this orbital tumor..

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