European Review for Medical and Pharmacological Sciences 2012; 16(4 Suppl): 90-94

Pleomorphic adenoma of the lacrimal gland: two clinical cases

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Abstract. – INTRODUCTION: Pleomorphic adenoma of the lacrimal gland is uncommon but it is the most common benign epithelial tumor of this gland. In the literature few cases have been reported in patients aged between 6 years and 80 years with a mean age of 39 years. A correct diagnosis and treatment is fundamental in order to avoid a relapse and sometimes their malignant transformation. An incisional biopsy is better to be avoided because it could injure the capsule, leading to dissemination of tumoral cells in the orbital tissues with a recurrence rate of 30% over 5 years.

AIM: This papers want to support the use of mini-invasive surgery for the treatment of orbital lesions when it is possible.

MATERIALS AND METHODS: We report two clinical cases of pleomorphic adenoma affecting the lacrimal gland treated with two different surgery approaches. The radiographic and photographic documentation of the patients was collected in the pre-and post-operatively. All patients underwent a CT scan and MRI.

CONCLUSIONS: This lesions requires a well-grounded clinical and therapeutic protocol to avoid the risk of malignant transformation or disease recurrence, very dangerous at this site. CT scan and MRI scan are very important to recognize different types of lesions involving the lacrimal gland and fossa. A mini-invasive surgery reduces hospitalization, risk of complications, surgical times and bleedings and guarantees an excellent functional and esthetic result when performed by a skilled surgeon.

Key Words:
Pleomorphic adenoma, Lacrimal gland, Epithelial tumor, Mini-invasive surgery.

Introduction

The lacrimal gland shares similar origins to the salivary glands. It is an exocrine gland and consists in an orbital lobe and in a palpebral lobe separated by the aponeurosis of the levator palpebrale superioris muscle. There is also the accessory lacrimal gland located in the lamina propria of conjunctiva. It helps producing the tear fluid, very important for the maintenance of a healthy ocular surface. In the lacrimal gland pathology a wide variety of causative factors are assumed to be possibly involved, like: aging, sex steroid hormone, neurotrophic factors, apoptosis, pituitary-depend factors, infections, smoke, autoimmune reaction.

Pleomorphic adenoma (PA) is the most common benign epithelial tumor of the lacrimal gland[1-5]. It is rare and risk factors are still unknown. It always develops by the deep orbital lobe, rarely from the palpebral lobe[6-8], extremely rarely from the accessory lacrimal gland[9], but it can be located everywhere there is glandular tissue. The authors present two cases of lacrimal gland PA treated by different surgery approaches, in the Department of Cranio Maxillo Facial Surgery, University of Rome "Sapienza".

Case 1

A 50 years-old female was referred to our Department with a four months history of diplopia and progressive loss of sight in the left eye. On the examination she had an exophthalmus, a deficiency in the left upward gaze (Figure 1A) and orbital dystopia. The conjunctiva was normal. She referred no pain and a history of surgery (in another hospital) for a left orbital mass defined as a fibrolipoma by its histology. After a careful examination, CT scan of the orbits was needed for a better comprehension of the problem and it showed: "…by the upper-external corner of the left orbit there is an oval solid inhomogeneous mass with regular margins that compresses and displaces anteriorly the ocular globe…" (Figure 2A). Thus, in order to better define the extension trough surrounding soft tissues, a MRI scan was performed and it showed: "…an expansive oval mass in the left orbit with regular margins that compresses and displaces anteriorly the ocular

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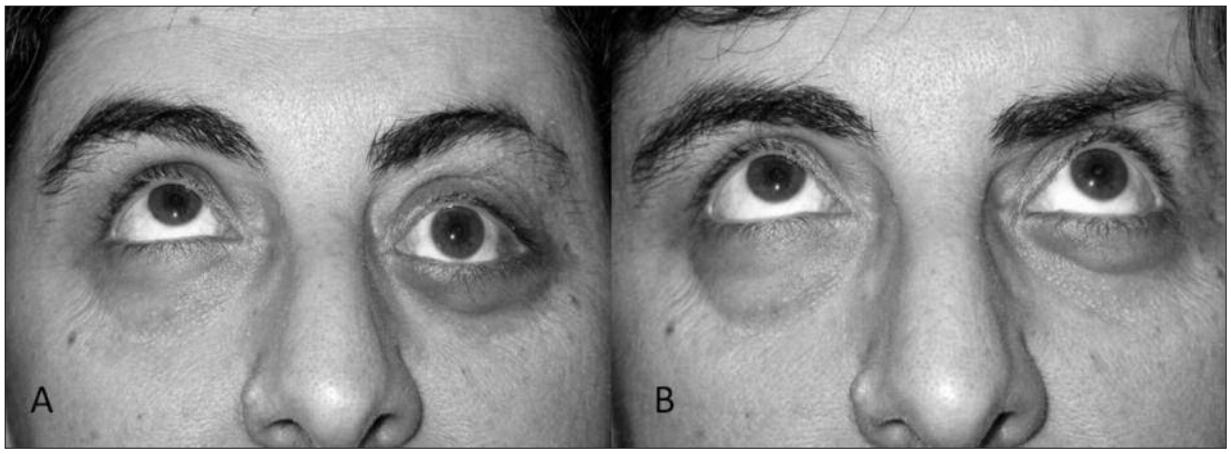

Figure 1. ***A,*** Patient with pleomorphic adenoma of the lacrimal gland showing shift of the leveling of the interpupillary line, conjunctival chemosis and limitation of the left eye in the upward movement. ***B,*** Patient three months postoperatively.

globe without evidence of local infiltration…". Because there wasn't any radiological and clinical evidence of malignancy and a total excision was possible, we didn't perform a pre-operative biopsy. By a coronal approach, after a partial resection of the temporal muscle, we exposed and performed an osteotomy on the left orbit lateral pillar. Once the oval mass was exposed, with its regular margins and a good cleavage plane, we completely removed it without drop the capsule. Surgery ended with the orbital pillar repositioning, fixed by plates and screws. Histopathology revealed that it was a pleomorfic adenoma (PA). After four days the patient was discharged. Following CT and MRI scans showed signs of the previous surgery without any evidence of recurrence (Figure 2B). There have been no signs of recurrence after a 7 years follow-up and she's still free of any ocular signs and symptoms (Figure 1B).

Case II

A 44 years-old woman presented at our Department with a short history (3 months) of left exophthalmos (Figure 3A). The patient was also visited by an oculist who performed ocular ultrasound that showed the presence of a left retrobulbar cystic mass. Ocular examination was unremarkable except for the left exophthalmos. The conjunctiva was normal. No pain was referred. So the patient underwent a MRI scan that revealed: "…an expansive mass on the upper-lateral region of the left eye

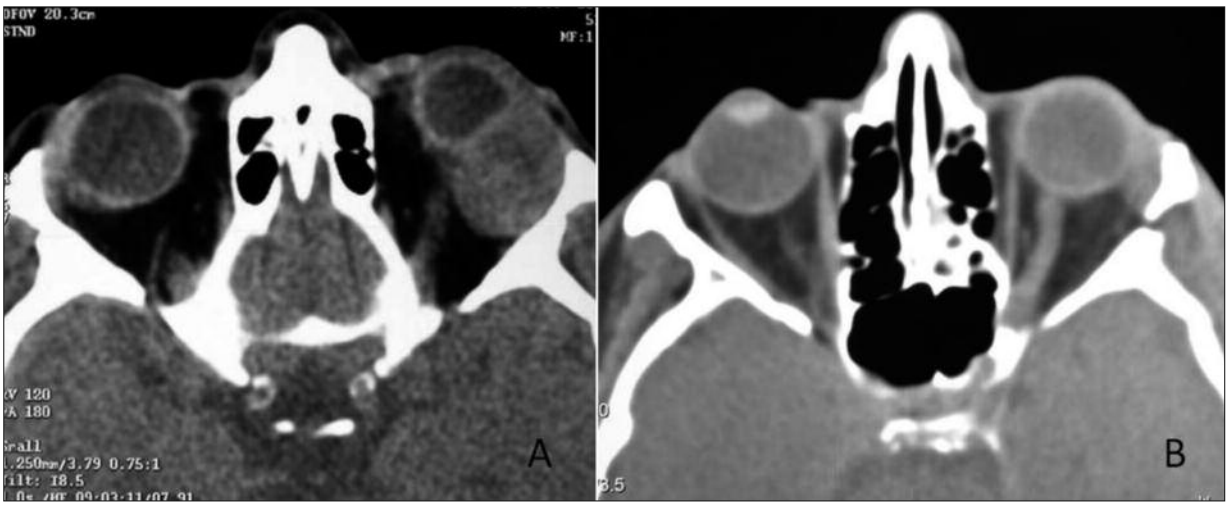

Figure 2. ***A,*** Axial computed tomography demonstrates an encapsuled mass localized in the upper external portion of the left orbital cavity. ***B,*** Postoperative CT scan didn't show any residual lesion.

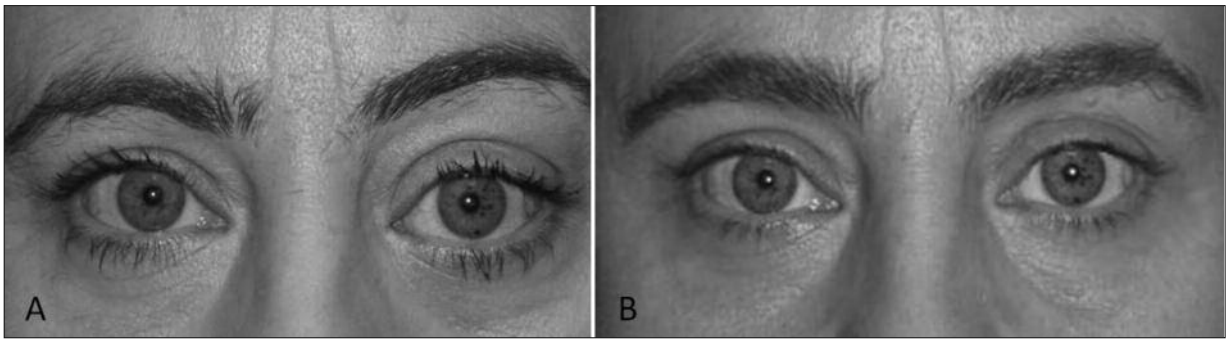

Figure 3. ***A,*** Preoperative photograph shows exophthalmus of the left eye. ***B,*** Postoperative photograph after four months: the scar is well hide in the eyelid fold.

with definite and regular margins, without any evidence of infiltration through the lateral and superior rectus muscles…". Moreover in order to better define the bone extension of the mass, we performed a CT scan that showed:"… an expansive mass on the upper-lateral region of the left eye (about 2.2 cm) without erosion of the orbital walls…" (Figure 4A).

Considering the imaging studies we chose to remove the lesion again without a pre-operative biopsy because the mass didn't show malignancy on the clinical and radiological exams as surrounding soft tissue invasion or bone erosion or irregular margins. So we performed a skin incision following the skin crease of the superior eyelid, then an osteotomy of the superior-external corner of the left orbit in order to have a better sight of the surgery field. We completely removed the mass without damaging the capsule and shortening both the surgery time and postoperative time of discharge (just 2 days!). Also we reached a good functional and esthetic result without any complications (Figure 3B). Macroscopically the tumor was 2.3 cm long and was well delimited within a thin capsule. A pathological diagnosis of PA of the lacrimal gland was made. After 3 years the patient remains free from disease as showed in post-operative CT and MRI scans (Figure 4B).

Discussion

Pleomorphic adenoma (PA) is the most common benign tumor of the lacrimal gland[10]. In the literature few cases have been reported in patients aged between 6 years[11] and 80 years[12] with a mean age of 39 years[13]. A correct diagnosis and treatment is fundamental in order to avoid a relapse and sometimes their malignant transformation. An incisional biopsy is better to be avoided because it could injure the capsule, leading to dissemination

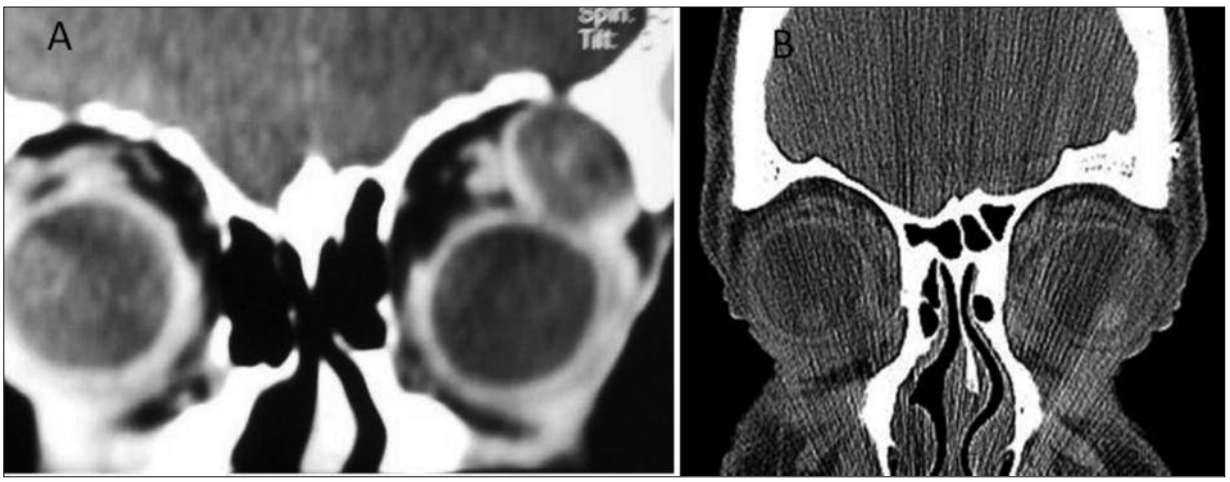

Figure 4. ***A,*** Coronal CT demonstrating a lobulated mass of the orbital lobe of the left lacrimal gland. ***B,*** Postoperative CT scan.

of tumoral cells in the orbital tissues with a recurrence rate of 30% over 5 years[5,14]. Though, it is very important performing a complete removal without injuring the capsule, the lacrimal artery and the lacrimal nerve, a branch of the ophtalmic division of the trigeminal nerve. The diagnosis is achieved by definitive histological examination after the excision of the mass[16].

About the surgical approach for orbital lesions, it varies depending on the site of origin, the extension and the histopathological diagnosis (when it is indicate and possible). In 1996 Rubin et al[17]described different approaches for the treatment of orbital disease by ophthalmologists, neurosurgeons, and maxillofacial surgeons. Seema et al[18] treated 32 cases of PA of the lacrimal gland with anterolateral orbitotomy through a Stallard-Wright incision and removed the tumor along with the periosteum and a rim of normal tissue.

We prefer, when possible, not to use these kind of complex surgical approaches that raise the risk of post-operative complications, prolonge the hospitalization with a mediocre esthetic result and a long surgical time. Porter et al[19], like us in the second case, excised en bloc a PA of the palpebral lobe of the lacrimal gland with a superior skin crease incision and the patient had no complications and remains asymptomatic at the 8 months follow-up. Clinically PA presents as a well circumscribed, slowgrowing, painless swelling mass that can cause ptosis, proptosis, exophthalmus, enophthalmus, orbital distopy, diplopia and restricted eye motility[20]. In our first case the patient presented with four months history of progressive loss of vision and diplopia. In the second case the only sign was exophthalmus. We both treated them by tumor excision without previous biopsy to avoid recurrences and malignant transformations. We also performed pre-operative CT and MRI scans in order to better define the lesion. CT scan is very important because its high sensibility for the presence of calcification and bony erosion, sclerosis and destruction. A disadvantage of CT is the inevitable exposure of the lens to irradiation. Magnetic resonance is superior in delineating the extent of soft tissue involvement. Disadvantages are the lower sensitivity for the presence of calcification, the imaging of the bone and the long image acquisition times.

In our opinion, in order to perform an appropriate approach for the treatment of these lesions, is fundamental a careful clinical and radiological study.

Conclusions

This paper aims to support the use of mini-invasive surgery for the treatment of orbital lesions when it is possible. With the advent of brain CT and MR scans, the orbital tumors are more easily disclosed and characterized. These studies are very important to recognize different types of lesions involving the lacrimal gland and fossa, but it's very difficult to differentiate each specific disease on the basis of image characteristics alone. A careful clinical evaluation and a pathologic study (when it is indicate and possible) are needed.

In the first case we performed an invasive surgical approach in order to have a wide vision of the operating field and avoiding a recurrence or a malignant trasformation, because the patient had already been treated. In the second case after a careful clinical and radiological study, we performed miniinvasive approach with a short surgery, completely removing the tumor and without intra and postoperative complications and a short hospitalization (2 days). The functional and esthetical results were very good. Therefore, after an appropriate clinical and radiological study, we'd like to promote, when possible, a mini-invasive surgery, which is better tolerated, shortens the hospitalization, reduces the risk of complications, surgical times and bleedings and guarantees an excellent functional and esthetic result when performed by a skilled surgeon.

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