In 1978, Font and Gamel presented follow-up data of 265 lacrimal gland lesions of which 136 (51%) had been classified as benign lacrimal gland pleomorphic adenoma (LGPA), 7 as malignant LGPA arising in LGPA, and 120 as other malignant lacrimal gland tumours. Follow-up (mean, 12.8 years) was available of 113 cases with benign LGPA. The average recurrence rate in this series was 13%. Interestingly, it was only 3% in the group in whom the tumor had been excised intact without a prior biopsy. When a biopsy had been taken, the recurrence rate was 32%, and when the pseudocapsule of the tumor had been damaged during surgery it was 21%. In the group in which a recurrence was excised, the lesion recurred again in 70%. These data thus made a strong case in favor of primary total removal of LGPA without a biopsy and with intact pseudocapsule.

Although, at first glance, this seems a logical conclusion, questions arise. No data are presented regarding the number of cases in which the tumor capsule was (further) damaged during tumor excision in the group who underwent a biopsy. No details are given regarding the applied surgical or biopsy techniques. It is unclear if the biopsy channel was excised. It is surprising that a biopsy is related to a higher recurrence rate then violation of the pseudocapsule during surgery.

In 1979, Wright and co-workers published data on a series of 40 patients with lacrimal gland tumors, of whom 20 suffered from LGPA and 20 of a primary malignant epithelial tumor. In 1979, Wright and co-workers published data on a series of 40 patients with lacrimal gland tumors, of whom 20 underwent a biopsy. No details are given regarding the applied surgical or biopsy techniques. It is unclear if the number of cases in which the tumor capsule was (further) damaged during tumor excision in the group who underwent a biopsy. No details are given regarding the applied surgical or biopsy techniques. It is unclear if the biopsy channel was excised. It is surprising that a biopsy is related to a higher recurrence rate than violation of the pseudocapsule during surgery.

In 1994, Henderson reported on 25 cases of LGPA. In 2009, Henderson reported on 25 cases of LGPA. In two cases, the tumor capsule was not intact on histological attenuation of the capsule, 9 underwent an open biopsy, 5 had preoperative breach of the pseudocapsule and 5 were operated after previous incomplete excision. During follow-up there was one recurrence, in a patient in whom a biopsy had been taken. The authors conclude that incomplete excision of LGPA predisposes for recurrence. While this conclusion suggests that, consequently, a biopsy should be avoided, the latter conclusion is not substantiated by the presented data.

In a 2009 paper, Rose stated the open biopsy is associated with a 10% chance of tumor recurrence, and that it would be foolish to biopsy a lesion that shows clinical and imaging characteristics of a LGPA. This 10% recurrence incidence proves based on the study described above, in which one out of 72 patients suffered from a recurrence. This patient, as well as nine others, had undergone an open biopsy, 5 had preoperative breach of the pseudocapsule and 5 were operated after previous incomplete excision. During follow-up there was one recurrence, in a patient in whom a biopsy had been taken. The authors conclude that incomplete excision of LGPA predisposes for recurrence.

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In 1997 Currie and Rose published another series of 133 patients surgically treated for LGPA. In 72 patients, follow-up was at least five years. In the latter group, intact excision had been performed in 46 patients, 7 showed histological attenuation of the capsule, 9 underwent an open biopsy, 5 had preoperative breach of the pseudocapsule and 5 were operated after previous incomplete excision. During follow-up there was one recurrence, in a patient in whom a biopsy had been taken. The authors conclude that incomplete excision of LGPA predisposes for recurrence. While this conclusion suggests that, consequently, a biopsy should be avoided, the latter conclusion is not substantiated by the presented data.

In a 2009 paper, Rose stated the open biopsy is associated with a 10% chance of tumor recurrence, and that it would be foolish to biopsy a lesion that shows clinical and imaging characteristics of a LGPA. This 10% recurrence incidence proves based on the study described above, in which one out of 72 patients suffered from a recurrence. This patient, as well as nine others, had undergone an open biopsy prior to excision. This extrapolation of one recurrence in a subset of nine patients to a 10% chance of recurrence after biopsy in a next paper lacks any statistical justification. The author also remarked that “with rare exceptions, benign tumors of the lacrimal gland are pleomorphic adenomas”. In this sentence, the word “epithelial” is missing. Furthermore, this statement is clinically not very helpful. Lymphocytic infiltration of the lacrimal gland or malignant lacrimal gland tumors are not always easily distinguished from a LGPA and even experienced orbitologists will not adequately diagnose LGPA in up to 22% of cases.

In 1994, Henderson reported on 25 cases of LGPA. In two cases, the tumor capsule was not intact on histological examination. In two other cases, the capsule ruptured during surgery. One other patient had piecemeal removal due to tumor necrosis, and in one other an open biopsy had been performed. In the whole group, no recurrences were seen after a follow-up ranging from 8 to 15 years.

Recently, Lai and co-workers reviewed the literature to find out if what evidence supports the claim that a biopsy should be avoided in LGPA. They concluded that data regarding the recurrence rate after an open biopsy are contaminated by confounding factors, such as incomplete tumor removal or possible spillage of tumor cells, to such extent that no conclusions can be drawn regarding the relation between an open biopsy of a LGPA and the risk of tumor recurrence.

Literature data do however suggest that incomplete excision of LGPA is associated with a higher recurrence rate as compared to complete removal. While this provides some support in favor of total removal of a LGPA without biopsy, the disadvantages of subtotal excision must also be weighed. As stated above, excision of the orbital lobe of the lacrimal gland will cause side-effects in more than 25% of cases. This percentage does not include any morbidity associated with the lateral orbitotomy procedure. Without biopsy, the diagnosis LGPA will be wrong in 15-22% of cases. Consequently, these subjects will either be overtreated or undertreated. Although it is unclear to what extent initial undertreatment affects survival in patients with a malignant lacrimal gland tumor we may assume that, by the same token, radical excision of these tumors constitutes the preferred treatment.

The 32% recurrence rate after biopsy of LGPA, as described by Font and Gamel in 1978 has not been reported since. It is, therefore, likely that their figure was based on inadequate (contaminated?) data. Nevertheless, the
opinion that LGPA should not be biopsied was repeated in literature many times since, and each time without adequate supporting data. Unfortunately, the statement that LGPA should not be biopsied is circular and hence illogical. You don’t need to biopsy a lesion if you know what it is, but you only know for sure what it is after you have taken a biopsy. Instead of an open biopsy, a fine needle aspiration biopsy (FNAB) can be used. Two authors described good reliability of FNAB in orbital mass lesions, especially when a combination of (immuno)histology and immunocytochemistry is used. These series, however, provide no precise data regarding how well FNAB will differentiate between LGPA and other lacrimal gland lesions.

In conclusion, authorities may present strong opinions that are not based on adequate data but that nevertheless create a mental box from which it proves difficult to escape, for themselves as well as for others. The assumption that LGPA should not be biopsied provides an excellent example of this mechanism. The application of FNAB is promising, but more precise data on its reliability in lacrimal gland lesions are needed.

References