



## LACRIMAL GLAND ADENOMA IN A SHEEP

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### Summary

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The lacrimal gland is a diamond-shaped, tubuloalveolar gland that secretes the serous component of tears. A four-year-old female crossbreed sheep suffering from left eye protrusion was referred to a Veterinary Hospital. Ophthalmic examination revealed epiphora, superficial ulcerative keratitis, corneal edema and neovascularisation. Moreover, ultrasound examination showed a large heterogeneous mass with variable reflectivity in the intraconal and extraconal spaces. Grossly, a 2.5×1.5×0.5 cm oval firm grayish mass was observed. Histopathologically, the mass was composed mainly by tubules with two cell types including cuboidal luminal epithelial cells and peripheral myoepithelial cells. The tubular structures were separated by proliferating myoepithelial cells. Mitotic figures, cellular pleomorphism and atypia were not seen. Immunohistochemically, most of the luminal epithelial cells showed an immunopositive reaction with a cytokeratin (AE1/AE3) marker. On the basis of these findings, the mass was diagnosed as a lacrimal gland adenoma.

**Key words:** histopathology, immunohistochemistry, lacrimal gland, neoplastic mass, sheep

The lacrimal gland is a specialised serous, diamond-shaped gland located in the dorso-lateral aspect of the globe lying within the periorbital space with 15–20 small ducts opening from it into the superior conjunctival fornix. Histologically, the gland is of the serous tubuloalveolar compound type (Gargiulo *et al.*, 1999; Samuelson, 2007). The lacrimal gland is a multilobular tissue composed of acinar, ductal, and myoepithelial cells. Sheep lacri-

mal glands are mixed glands, consisting of tubulo-acinar units that are succeeded by ducts of simple morphology (Tosaka, 1991; Gargiulo *et al.*, 1999; Peiffer *et al.*, 1999). This gland secretes the serous component of tears, which then flows into canals that lead to the lacrimal sac (Wilcock, 2007). Lacrimal gland tumours are slowly growing tumours and usually, arise on the margin of the eyelid, but often extend into the eyelid itself where they pre-

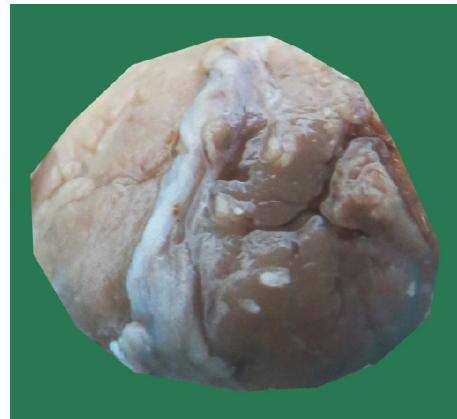
sent as a bulging of the conjunctiva (Hirayama *et al.*, 2000).

In humans, the most frequent primary epithelial tumours are benign pleomorphic adenomas (Kohli *et al.*, 2011). In animals, tumours of lacrimal gland have been reported rarely and there are only a few reports of benign or malignant primary growths (Peiffer *et al.*, 1999; Wang *et al.*, 2001; Wilcock, 2007; Kohli *et al.*, 2011). To the best of our knowledge, to date, there is no report about lacrimal gland adenoma in sheep. Therefore, the present report describes the clinicopathological and immunohistochemical findings of a rare case of lacrimal gland adenoma in an adult ewe.

**Case presentation.** A four-year-old female crossbred sheep suffering from left eye protrusion with a unilateral enlarged nonulcerated mass in the dorsolateral aspect of the left orbit was referred to the Veterinary Hospital at the School of Veterinary Medicine, Shiraz University. The swelling of the left eye and eyelid had been noticed by the owner, two months prior to presentation. Clinical signs were epiphora, ocular discharge, keratoconjunctivitis, thickening and partly coarseness of the upper eyelid. The skin was normal, thin, elastic, and movable over the mass, which protruded outward or extended into the eyelid. On ophthalmic examination, conjunctival congestion, epiphora, superficial ulcerative keratitis, mild corneal oedema, and corneal neovascularisation were diagnosed. Moreover, ultrasound examination of the left orbit revealed a large heterogeneous mass with variable reflectivity in the intraconal and extraconal spaces, especially in the medial and superior orbit. Posterior chamber, iris, and lens were healthy, but the anterior chamber of the eye was abnormal and the cornea was affected by the external tissue.

The optic nerve shadow could be seen separately from the lesion anteriorly. The mass on the eyelid was surgically excised under local anaesthesia. Appropriate samples were fixed in 10% neutral buffered formalin, dehydrated in graded ethanol, cleared in xylene, and embedded in paraffin wax. Sections of 5 µm thicknesses were stained with haematoxylin and eosin (H&E) and studied microscopically. For the immunohistochemical study, cytokeratin antibody (AE1/AE3; Biogenex, USA) was used as the primary antibody.

Grossly, after surgical excision, a 2.5×1.5×0.5 cm oval firm grayish mass was identified in the inner part of the left upper eyelid (dorsolateral to the left globe). The mass was encapsulated and relatively distinct from the surrounding tissues. The cut surface revealed a firm, grayish multilobulated tissue (Fig. 1).



**Fig. 1.** Lacrimal gland adenoma. An oval gray neoplastic mass formation 2.5×1.5×0.5 cm of size.

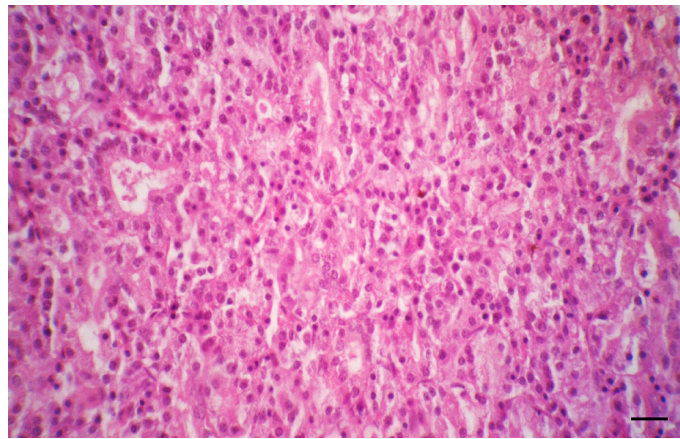
Histopathologically, the mass was surrounded by a capsule of thin connective tissue and composed mainly of tubules with two cell types. These cells consisted of cuboidal luminal epithelial cells with small round dense basally located nuclei

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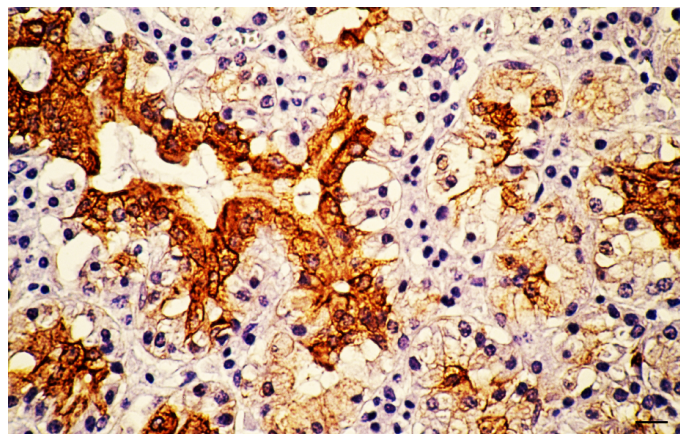
and peripheral myoepithelial cells. The tubular structures were separated by proliferating myoepithelial and mononuclear inflammatory cells. However, the results revealed no mitotic figures, cellular pleomorphism and other malignancy characteristics (Fig. 2). Further, the neoplastic mass was not observed to be infiltrated into the globe and the surrounding tissues. Immunohistochemically, most of the luminal epithelial cells showed immunopositive reactions with the cytokeratin (AE1/AE3) marker (Fig. 3).

On the basis of histopathological and immunohistochemical findings, the mass was diagnosed as a lacrimal gland adenoma.

Glands of the eyelid consist of sebaceous glands such as Meibomian (tarsal) and Zeis glands that open to hair follicles and Moll's glands that have modified sweat gland characteristics (Goldschmidt & Shofer, 1992; Dellman & Eurell, 1998; Johnson *et al.*, 1999; McGavin *et al.*, 2001). Tumours in the orbit itself are a serious diagnostic and therapeutic challenge.



**Fig. 2.** Small to large of tubular cells are separated from each other by proliferating myoepithelial and inflammatory cells (H&E, bar: 100  $\mu$ m).



**Fig. 3.** The neoplastic cells show positive staining for cytokeratin AE1/AE3 (IHC, bar: 25  $\mu$ m).

Diagnosis is constrained by the lack of effective simple imaging methods, and therapy is a challenge because of the inaccessibility of the retrobulbar structures. The advent of better-quality ultrasound facilities and in particular MRI and CT diagnostics has enormously improved the imaging of the orbital structures (Lavach & Severin, 1977; Miesner *et al.*, 2009). In humans, lacrimal gland tumours represent 5% to 10% of orbital lesions, and the epithelial proportion, as shown in the literature, ranges from 23% to 70% of biopsied cases (Reese, 1956; Ni *et al.*, 1992; Font *et al.*, 1998; Rootman, 2003).

For classification of lacrimal gland tumours, in 2006, the Armed Forces Institute of Pathology (AFIP) monograph showed an extensive classification based on the 1992 World Health Organization (WHO) classification of salivary gland tumours. Therefore, it is clear that the salivary gland classification has filtered into the lacrimal gland literature, which has described many tumours analogous to their salivary gland counterparts, including ductal carcinoma, acinic cell carcinoma, primary squamous cell carcinoma, mucoepidermoid carcinoma, oncocytic carcinoma, polymorphous low-grade adenocarcinoma, myoepithelial carcinoma, lymphoepithelial carcinoma, epithelial-myoepithelial carcinoma, cystadenocarcinoma, primary sebaceous adenocarcinoma, basal cell adenocarcinoma, oncocytoma, cystadenoma, and myoepithelioma (Head, 1990).

The two main types of lacrimal gland tumours in humans are pleomorphic adenoma and adenoid cystic carcinoma. In dogs, neoplasms of the lacrimal gland are rare, but most primary epithelial tumours are reported to be lacrimal gland adenocarcinomas (Head, 1990). Malignant tumours of the lacrimal gland include ade-

nocystic carcinomas, squamous cell carcinomas, mixed malignant tumors, adenocarcinomas, and lymphomas. They may cause ocular irritation or discomfort leading to ocular discharge and blepharospasm, and may ultimately lead to corneal damage and ulceration. Benign lacrimal gland tumours in human beings are mostly pleomorphic adenomas which are non-invasive and follow a benign course (Brown, 2002). Similar to the present study, a case of pleomorphic adenoma of the lacrimal gland was reported in a dog with an immunopositive reaction against antihuman keratin/cytokeratin (AE1/AE3) in the luminal epithelial cells (Hirayama, 2000).

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