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CASE REPORT

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Dacryops with dacryolithiasis in a dog

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Abstract

Background: A 10-year-old castrated male Maltese dog was presented with chronic swelling that had been present for at least 5 years in the medial canthus of the right eye (OD).

Objectives: To describe the treatment outcome of dacryops with dacryolithiasis.

Methods: Bilateral patency of the nasolacrimal system was confirmed by flushing of both upper and lower puncta. Ocular ultrasonography revealed a well-defined, oval-shaped, heterogeneous mass with several hyperechoic foci. Dacryocystorhinography revealed no connection between the mass and lacrimal canaliculus. Gentle blunt dissection of the fibrous connective tissue around the cystic mass was performed. The mass was removed, which intraluminally contained multiple calculi.

Results: Histopathologically, the cystic structure was lined by simple cuboidal epithelium and surrounded by smooth muscle actin positive myoepithelial cells consistent with dacryops derived from the lacrimal glandular ductal system. In addition, several spherical basophilic minerals were observed in the lumen, which were identified as dacryoliths.

Conclusion: Surgical removal of this dacryops with dacryolithiasis was curative without recurrence after four months.

KEYWORDS
dacryolithiasis, dacryops, ectopic lacrimal gland, lacrimal system, Maltese, nasolacrimal cyst

1 | INTRODUCTION

A lacrimal cyst (dacryops) is a rare cystic dilation of the lacrimal ducts or gland that may originate from the orbital lacrimal gland or the nictitans gland (Giuliano, 2013; Leiva & Gimenez, 2018; Pickett, 2018). The cause of dayhops is usually unknown (Pickett, 2018), but developmental defects, foreign body injury, blunt trauma or inflammation affecting the ducts are thought to be possible causes (Giuliano, 2013). Frequent reports in young basset hounds and Labrador retrievers (Ota et al., 2009; Playter & Adams, 1977) suggest a congenital condition and a presumed predisposition in those breeds (Giuliano, 2013; Leiva & Gimenez, 2018; Ota et al., 2009; Pickett, 2018). Dacryops have also been reported in the German shepherd dog (Cullen & Grahn, 2003), Neapolitan mastiff (Delgado, 2013), golden retriever (Lamagna et al., 2012) and cats (Maggio, 2020; Sritrakoon et al., 2016). A dacryops is suspected based on clinical signs and location (Pickett, 2018). They are confirmed by fine-needle aspiration of the cystic fluid and cytology, dacryocystorhinography, rhinoscopy, surgical biopsy and surgical

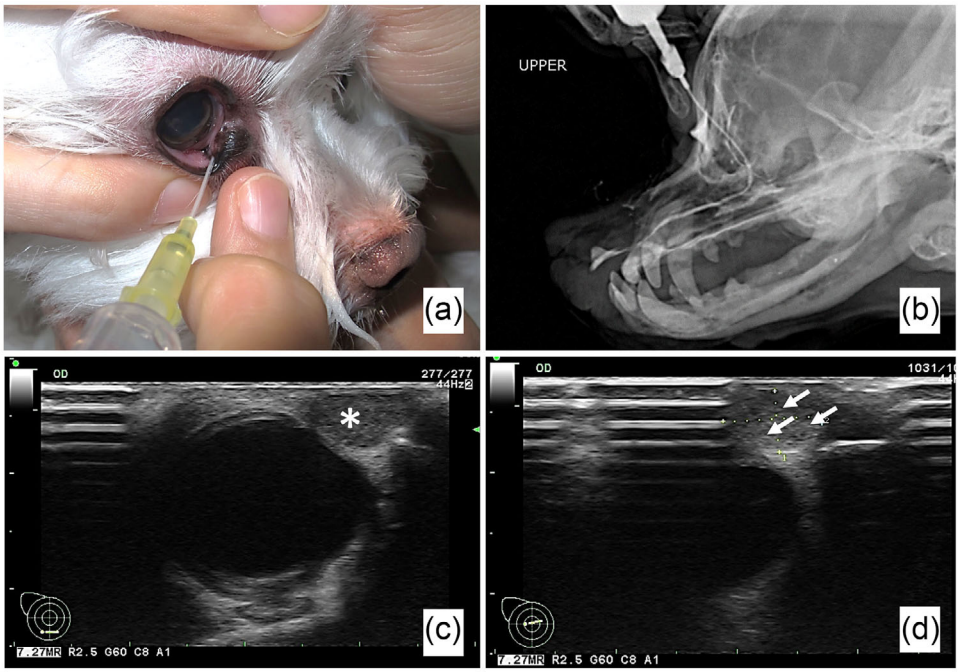

FIGURE 1 Diagnostic approaches used. (a) Nasolacrimal flushing of the lower nasolacrimal puncta of the right eye; (b) contrast dacryocystorhinography of the nasolacrimal duct showed no connection to the mass; (c) ocular ultrasonography showed a well-defined, oval-shaped, heterogeneous mass (asterisk); (d) the cyst contained several hyperechoic foci (arrows)

exploratory excision of the pathologic tissue, which is also the treatment of choice for this condition (Leiva & Gimenez, 2018; Pickett, 2018).

Dacryoliths, or nasolacrimal calculi, are a concretion within the nasolacrimal system and are uncommon in animals. Only three reports exist in the veterinary literature: one in the distal nasolacrimal duct of a horse, one contained in canaliculops (cyst of canalicular origin) of a Labrador retriever, and one in both orbitae and nasolacrimal ducts of a snow leopard (Cassotis & Schiffman, 2006; Malho et al., 2013; Wiesner et al., 2019). We describe a case of dacryoliths contained in a dacryops in a Maltese dog. Nasolacrimal flushing, ocular ultrasound and dacryocystorhinography were performed to confirm the diagnosis and plan the surgical procedure.

2 | CASE HISTORY

A 10-year-old castrated male Maltese dog presented with enlargement of the medial canthal mass in the right eye (Oculus Dexter [OD]); Supplementary Figure S1a ([Supplementary Figure S1b is the left eye]). The mass was not painful and had been noted by the owner for at least 5 years but had been rapidly growing recently. Ophthalmic examinations were performed by a board-certified veterinary ophthalmologist (Asian College of Veterinary Ophthalmologists). The Schirmer tear test-1 (Schering Plough) values were 17 mm/min in both eyes (Oculi Uterque [OU]). No epiphora was observed. Intraocular pressure measured using the rebound tonometer (TonoVet; iCare) was 14 mmHg OD and 17 mmHg in the left eye. Neuro-ophthalmic examinations were normal OU. Slit-lamp biomicroscopy (SL-D7; Topcon) revealed nuclear sclerosis and focal cataracts in both lenses. Indirect ophthalmoscopy showed retinal degeneration and retinal blood vessel attenuation OU. The dog was able to negotiate the maze test under both photopic and scotopic conditions.

Nasolacrimal flushing confirmed the patency of the nasolacrimal duct OU with slight resistance on the inferior lacrimal punctum OD (Figure 1a). Contrast dacryocyctorhinography revealed normal anatomy of the nasolacrimal duct with no connection to the mass (Figure 1b). B-mode ocular ultrasonography revealed a well-defined oval-shaped cyst with multifocal heterogeneous echogenicities within its wall (Figure 1c), with a swirling motion of the internal contents consisting of several hyperechoic foci (Figure 1d). The size of the cyst was 0.51 × 0.87 cm. Because the cyst structure was clearly identified through soft tissue ultrasound and the X-ray showed no bone anomalies, further imaging diagnostics, such as computed tomography (CT) imaging, were not performed.

Surgery was performed under routine general anaesthesia. The lower lacrimal punctum OD was cannulated with a 24-gauge intravenous catheter to protect the canaliculus located nearby, and a skin incision was made parallel to the lower eyelid margin (Figure 2a). The cyst was inadvertently ruptured during the surrounding tissue dissections, and several small stones were identified inside the cyst lumen along with turbid fluid (Figure 2b and Supplementary Video File). These stones were light cream in colour, with sizes ranging from 0.1 to 1 mm in diameter. The intraluminal fluid was submitted for cytology and culture/antibiotic susceptibility testing. Although the cyst wall was thin enough to rupture, the cyst was well-separated from the

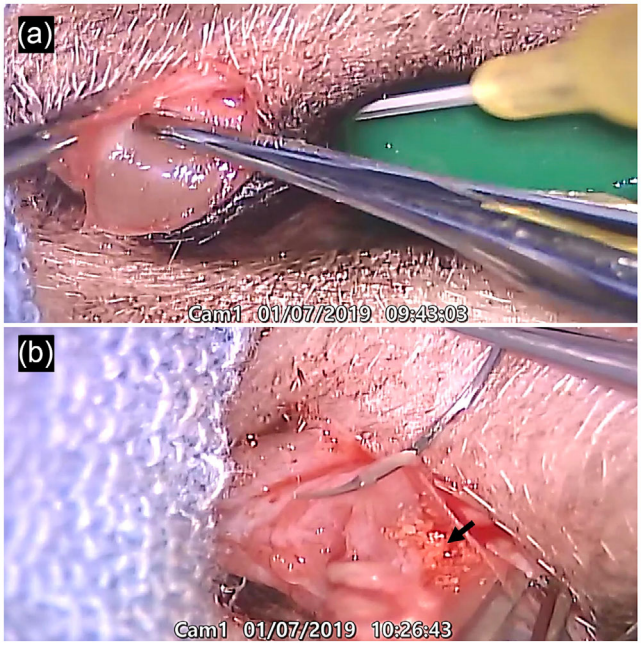

FIGURE 2 Intraoperative photograph of cyst dissection. (a) Blunt dissection of the ventromedial eyelid revealed a cyst (dacryops). (b) The cyst contained several small-sized stones (arrow)

surrounding tissue and was able to be completely removed (Supplementary Video File). The subcutaneous tissue was sutured with 8–0 polyglactin (Vicryl; Ethicon), and the skin was closed using 6–0 polyglactin (Vicryl). After the surgery, flushing of the nasolacrimal duct was performed through the upper and lower puncta to confirm the patency of the canaliculi. The cyst was submitted for routine histopathological examination. Chemical analysis of the dacryoliths was not performed due to the owner's financial constraints.

Oral amoxicillin/clavulanic acid 12.5 mg/kg q12 h (Clavamox; Pfizer) and carprofen 2.2 mg/kg q12 h (Rimadyl; Zoetis) were prescribed for 5 days. Topical levofloxacin 0.5% (Cravit; Santen) Quarter In Die and artificial tears (Refresh Plus; Allergan) prn were also prescribed OD. Cytology of the fluid content revealed few monocytes and amorphous basophilic material with no bacteria. In addition, culture/antibiotic susceptibility testing showed no growth, suggesting that no infections were involved. Histopathologically, the cyst was lined by simple cuboidal epithelium and contained a few basophilic mineral-like materials in the lumen, identified as dacryoliths (Figure 3a,b). Immunohistochemistry revealed smooth muscle actin (SMA)-positive cells in the outer layer near the ductal epithelium, indicating the presence of myoepithelial cells, which was consistent with the glandular duct (Figure 4a). SMA-positive blood vessels on the tissue around the cyst acted as a positive control (Figure 4b).

Follow-up examination 11 days post surgery showed adequate healing of the periocular surgical site, and the stitches were removed. At 18 days postoperatively, the swelling at the surgical site subsided, and the appearance of the lower eyelid was normalised (Supplementary Figure S1c). Jones Test 1 was performed and was positive. No epiphora or any other abnormalities were detected on ophthalmic examinations 4 months postoperatively (Supplementary Figure S1d). No recurrence of the cyst was observed during the 4-month follow-up.

3 | DISCUSSION

Dacryops, although rare, have been mostly reported in young dogs aged 3.5 months to 1 year (Cullen & Grahn, 2003; Delgado, 2013; Lamagna et al., 2012; Ota et al., 2009; Playter & Adams, 1977). Two reports of dacryops in cats also occurred in 1-year-old and younger cats (Maggio, 2020; Sritrakoon et al., 2016). In fact, dacryops is generally considered a congenital disorder in animals (Giuliano, 2013; Leiva & Gimenez, 2018), hence the diagnosis at a young age. In the present case, the dog presented to the veterinarian at the age of 10 years, with the chronic medial canthal mass being present for at least the past 5 years. Recently, dacryops cases in dogs 3–11 years of age were reported to be accompanied by maxillary bone defects (Steinmetz et al., 2021).

Compared to dacryops, dacryolithiasis is even more rare in animals, with only one case in a 12-year-old Paso Fino mare (Cassotis & Schiffman, 2006), one case in a 4-year-old Labrador retriever

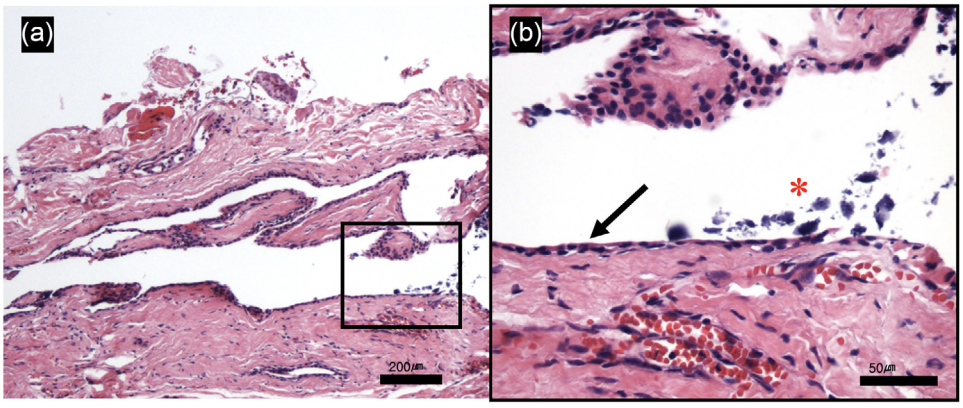

FIGURE 3 Histopathology of the cyst in the medial canthus of the right eye. The cyst wall was lined by simple cuboidal epithelium (arrow). A few basophilic mineral-like materials were present inside the lumen (asterisk). Hematoxylin and Eosin staining. Bar (a) = 200 μm; (b) = 50 μm.

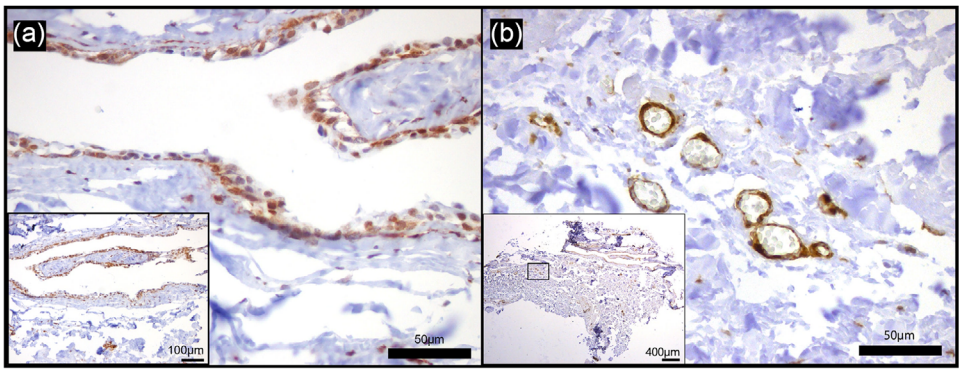

FIGURE 4 Immunohistochemistry of the cyst from the medial canthus of the right eye. (a) The cyst was surrounded by smooth muscle actin (SMA)-positive cells in the outer layer of the ductal epithelium. (b) SMA-positive blood vessels on the tissue around the cyst, serving as a positive control.

(Malho et al., 2013), and one case in a juvenile snow leopard (*Uncia uncia*; Wiesner et al., 2019), making our patient the second canine case. However, the cyst in the Labrador originated from the lacrimal canaliculus (canaliculops), while in the present case, it originated from the glandular tissue and/or glandular duct (dacryops). Meanwhile, previous reports on canine dacryops did not include the formation of dacryoliths (Cullen & Grahn, 2003; Delgado, 2013; Lamagna et al., 2012; Ota et al., 2009; Playter & Adams, 1977; Steinmetz et al., 2021). In humans, dacryolithiasis is also considered rare. However, the presence of dacryoliths on both dacryops and a nasolacrimal cyst from the lacrimal sac or canaliculus origin has been reported in humans (Iliadelis et al., 1999; Mishra et al., 2017; Orhan et al., 1996; Salam et al., 2012).

In veterinary medicine, dacryolithiasis in a Paso Fino mare manifested as a calculus at the distal nasolacrimal duct accompanied by a history of chronic ocular and nasal discharge associated with bacterial infections (Cassotis & Schiffman, 2006). In the present case, the cytology and culture results suggested that no infections were involved. The dacryoliths in Labrador were also not associated with infections (Malho et al., 2013). In humans, dacryoliths have been reported in both acute and chronic dacryocystitis (Iliadelis et al., 1999; Orhan et al., 1996; Salam et al., 2012). The dacryoliths in the horse had a chronic history of ocular and nasal discharge for more than a year (Cassotis & Schiffman, 2006). Aside from chronic dacryocystitis, dacolith formation has been associated with intraluminal foreign bodies, eyelash niduses and stasis of secretions when infections are not involved (Cassotis & Schiffman, 2006; Iliadelis et al., 1999; Orhan et al., 1996). Considering the lack of infections and any identified foreign body or nidus, the chronic existence of the cyst in the present case might have resulted in dacolith formation. The stasis flow inside the dacryops for more than 5 years could have led to mineralisation.

Unlike the cyst of canaliculus origin, which is lined by stratified squamous epithelium, the present cyst was lined by simple cuboidal epithelium consistent with the cyst derived from lacrimal glandular and ductal tissues (Giuliano, 2013; Malho et al., 2013; Ota et al., 2009). In addition, the presence of myoepithelial cells around the cyst wall confirmed through immunohistochemistry for SMA suggests that the cyst originated from the lacrimal glandular duct and not from the canaliculus (Cullen & Grahn, 2003; Ota et al., 2009). However, since canine lacrimal tissue does not normally exist in the medial canthus, this is most likely derived from ectopic or choristomatous lacrimal tissue (Lamagna et al., 2012; Ota et al., 2009; Playter & Adams, 1977). Surgical removal of the cyst is the recommended treatment for the management of dacryops, as in this case, with the majority of cases being successful. Alternatively, an injection of a sclerosing agent following aspiration of the cyst contents using either tetracycline or 1% polidocanol can be performed (Stuckey et al., 2012; Zimmerman & Stefanacci, 2019). However, due to the possibility of conjunctival necrosis and severe periocular inflammation, it is not routinely recommended (Giuliano, 2013). In the current case, the dacryoliths within the dacryops might have affected the action of the sclerosing agent.

Some limitations identified in the current study were the lack of chemical analysis of the dacryoliths and the absence of CT images. Since bone anomalies cannot be excluded definitively by X-ray, the possibility of these dacryops accompanied by a bone defect remained. To the best of our knowledge, this is the first report of dacryops with dacryoliths in dogs. The chronic course of dacryops in this case may have played a role in the formation of dacryoliths. Surgical removal of the cyst was curative without recurrence 4 months after surgery.

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CONFLICT OF INTEREST

None of the authors of this article has a financial or personal relationship with other people or organisations that could inappropriately influence or bias the content of the paper.

ETHICS STATEMENT

This study was approved by the animal hospital and clinic board. Additionally, the animal's owner consented to the clinical treatment and the use of related information in the publication.

AUTHOR CONTRIBUTIONS

Conceptualisation, *formal analysis*, *investigation*, *writing–original draft*: Lina Susanti. *Supervision*, *validation*, *writing–review and editing*: Kangmoon Seo. *Conceptualisation*, *Investigation*, *Methodology*, *Supervision*, *Validation*, *Writing–review and editing*: Seonmi Kang.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

PEER REVIEW

The peer review history for this article is available at https://publons.com/publon/10.1002/vms3.853

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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