

CASE REPORT

Dacryops with extensive lacrimal and maxillary bone defects in four dogs

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Abstract

Objective: To describe and discuss ventromedial orbital lacrimal gland or duct cysts (dacryops) in dogs with extensive bone defects based on their symptoms, results of diagnostic imaging and histopathological examination, and therapy and discuss their potential origin based on the morphology.

Animals studied: Four dogs of different breeds, age, and sex were presented with a unilateral round, slow growing, indolent, and non-tender process ventromedial to the nasal canthus of the eye.

Procedures: Transverse computed tomography showed a low-density, non-contrast-enhancing cystic process ventromedial to the globe with extensive defects in the lacrimal and maxillary bones in all cases. The cystic character of the structure was confirmed by the aspiration of the brownish fluid without cellular and microbiological contents. For treatment, the cystic fluid was aspirated, and the sclerosing agent polidocanol was injected in three cases. Cystorhinostomy (nasal marsupialisation) was performed in one case as the first choice and in another case following failure of sclerotherapy. Histopathological examination of the cyst walls was performed in two cases and confirmed the diagnosis of dacryops.

Results: Follow-up between 2 and 18 months showed no recurrence and very good to excellent cosmetic results.

Conclusions: Aberrant lacrimal gland or duct tissue with secondary development of dacryops should be included in the differential diagnoses of ventromedial orbital cysts. Large cysts near the lacrimal drainage system with extensive bone defects in dogs should be treated by nasal marsupialization. Treatments such as evacuation of the cyst and inducing sclerosis (sclerotherapy) should be reserved for exceptional cases.

KEYWORDS

cystorhinostomy, dacryops, diagnostic imaging, dog, sclerotherapy, ventromedial orbital cysts

1 | INTRODUCTION

Fluid-filled lesions of the periorbital region are rare in humans and companion animals. Nevertheless, there are several

glandular and saccular structures in the periorbital region, which have the potential to produce a cystic lesion. The respective location of the cyst has been described as the most important clinical indicator of the tissue of origin.¹ As an

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example, a odontogenic parakeratinized cyst was reported to result in exophthalmos and palatine, maxillary, and zygomatic bone erosion in a dog.² Ventromedial lacrimal orbital cysts have been named inconsistently in veterinary literature: for example dacryops^{1,3–5} maxillary bone epithelial cyst,^{6,7} periorbital cyst with bone defect,⁸ orbito-nasal cyst,⁹ dysonotogenic cyst,¹⁰ naso-lacrimal duct cyst,¹¹ and periorbital epidermoid cyst.¹²

Orbital cysts close to the naso-lacrimal system but without any physical anatomical connection were described in some of these case reports, in dogs^{1,3,5,6,12} and a horse.⁴

The aims of the present study were to describe a special type of ventromedial orbital cyst independent of the naso-lacrimal duct (NLD) with extensive bone defects in dogs. We hereby present the symptoms, results of diagnostic imaging and histopathology, and therapy, as well we discuss the potential origin of the lesion based on morphology.

2 | MATERIALS AND METHODS

A 4-year-old male German pinscher (case 1), an 11-year-old male miniature dachshund (case 2), a 3-year-old male Bernese mountain dog (case 3), and a 10-year-old female mixed-breed dog (case 4) were presented with a unilateral round, slow-growing process medio-ventral to the nasal canthus of the eye. (Figure 1).

Clinical data were obtained by slit-lamp-biomicroscopy (SL 17, Kowa company Ltd.) (cases 1–4), control of the patency of the NLD by Jones test (cases 1–3), aspiration and cytologic and microbiologic analyses of the cyst contents (cases 1, 2 and 4). Further investigations performed were computed tomography (CT) (cases 1–4), testing the passing ability for drainage during the surgery and histopathological examination of the cystic wall (cases 3 and 4). The clinical data of the animals are summarized in Table 1.

All dogs were anaesthetized for CT and subsequent therapy. Anesthesia was induced intravenously with 0.5 mg/kg diazepam (Faustan®, Temmler Pharma) and 0.5 mg/kg levomepethadone in a fixed combination with 0.025 mg/kg fenipramide (L-Polamivet®, Intervet). Anesthesia was maintained via endotracheal intubation with 1% isoflurane (Isofluran CP, CP Pharma) dissolved in oxygen at a flow rate of 10 mL/kg/min.

In cases 1, 2, and 4, the cysts were tapped with a venous catheter (22 G, Vasofix®, Braun). The maximum possible amount of cystic fluid was aspirated after removing of the stylet. Thereafter, 1–2 mL (depending on cyst size) of the sclerosing agent polidocanol (Aethoxysklerol® 1%, Chemische Fabrik Kreussler & Co. GmbH) was injected.

Cystorhinostomy was performed in case 3 as the first choice and in case 4 following failure of sclerotherapy: First, a small silicone tube was passed down the lacrimal duct from the upper punctum lacrimale to protect and delineate the ductal system during the surgical procedure. Subsequently, a skin incision was made beginning 1 cm distal to the medial canthus about 4 cm in the caudoventral direction. Using the lateral approach, the fibrous tissue was bluntly and sharply dissected, the outer wall of the cyst was opened and the fluid was removed. Marsupialisation of the cyst to the nasal cavity was performed by complete resection of the inner cystic wall. Subsequently, the lateral fibrous and subcutaneous tissues were meticulously closed with Sultan's diagonal sutures using 3/0 polydioxanon (PDS®, Ethicon) to create adequate and firm coverage of the bone defect. The skin was closed with interrupted sutures using 4/0 polyamide (Ethilon®, Ethicon) (Figure 2).

Systemic robenacoxib 1 mg/kg (Onsior®, Elanco) was administered orally s.i.d for 3 days for analgesia postoperatively in cases 3 and 4 and in case 1 after rupture of the cystic wall. Amoxicillin-clavulanic acid 20 mg/kg (Synulox®, Zoetis) was administered orally b.i.d. for 10 days postoperatively in cases 3 and 4.

Follow-up was done clinically (in case 4) and by phone (in cases 1–3) and included assessment of recurrence of the cystic structure, cosmetic results, and the presence of lacrimal discharge.

3 | RESULTS

An overview of the clinical data, therapies, and outcomes is presented in Table 1.

The patients of the study were middle aged to old, dogs from different breeds and gender. Time between onset of the appearance of the cystic process and presentation of the animals to our hospital was more than 6 months in all cases.



FIGURE 1 Case 1–4: cystic process medio-ventral to the nasal canthus of the eye

TABLE 1 Clinical data, therapies, and outcomes of the four cases

| Case | 1 | 2 | 3 | 4 |
|--|---|---|---|--|
| Breed | German Pinscher | Miniature Dachshund | Bernese Mountain Dog | Mixed breed |
| Age (years) | 4 | 11 | 3 | 10 |
| Gender | Male | Male | Male | Female |
| Affected Orbita | Left | Left | Left | Right |
| Time between onset and presentation (months about) | 7 | 6 | 8 | >12 |
| Estimated diameter of the cystic process (cm) | 1.5 | 2.5 | 2.5 | 3 |
| Slit-lamp findings | Serous discharge | Serous discharge nucleosclerosis | Serous discharge | Serous discharge nucleosclerosis |
| Patency of the Naso-lacrimal duct (therapy ahead) | Jones 2 negative | Jones 1 and 2 negative | Jones 1 negative; during the operation confirmed by passability for drain | During the operation confirmed by passability for drain |
| Cystic fluid | Serous brown fluid | Serous brown fluid | Serous brown fluid | Serous brown fluid |
| Cystic fluid analyses | Some protein; small amount of hemosiderophages, lytic cells, some neutrophils | Some protein, some neutrophils | n.e. | Some protein, small amount of hemosiderophages, epithel cells, neutrophils, lymphocytes |
| Microbiologic analyses of the cystic fluid | Negative culture | Negative culture | n.e. | Negative culture |
| Diagnostic imaging | Computed tomography (CT) | CT | CT | CT |
| Therapy | Two times: Aspiration of the cyst fluid and instillation of 1 ml polidocanol | Aspiration of the cyst fluid and instillation of 1 ml polidocanol | Cystorhinostomy | First: Aspiration of the cyst fluid and instillation of 2 ml polidocanol Second: Cystorhinostomy |
| Aftercare | Only after the first injection: robenacoxib 1 mg/kg sid for 3 days | None | Amoxicillin-clavulanic acid 20 mg/kg bid for 10 days robenacoxib 1 mg/kg sid for 3 days | Amoxicillin-clavulanic acid 20 mg/kg bid for 10 days robenacoxib 1 mg/kg sid for 3 days |
| Histopathology cyst wall | n.e. | n.e. | Attenuated epithelium, rarely a double layer of cuboidal cells could be appreciated | Attenuated epithelium, rarely a double layer of cuboidal cells could be appreciated; occasionally epithelial lining with replacement by granulation tissue |
| Follow-up (months) | 18 | 2 | 5 | 2 |
| Recurrence after last therapy | No | No | No | No |

(Continues)

TABLE 1 (Continued)

| Case | 1 | 2 | 3 | 4 |
|------------------------------------|-----------|-----------------------------|-----------------------------|--------------------------|
| Lacrimal discharge at last control | None | Very little mucoid fluid | Very little mucoid fluid | Very little mucoid fluid |
| Cosmetic result at last control | Excellent | Very good Small indentation | Very good Small indentation | Excellent |

Abbreviation: n.e., not examined.

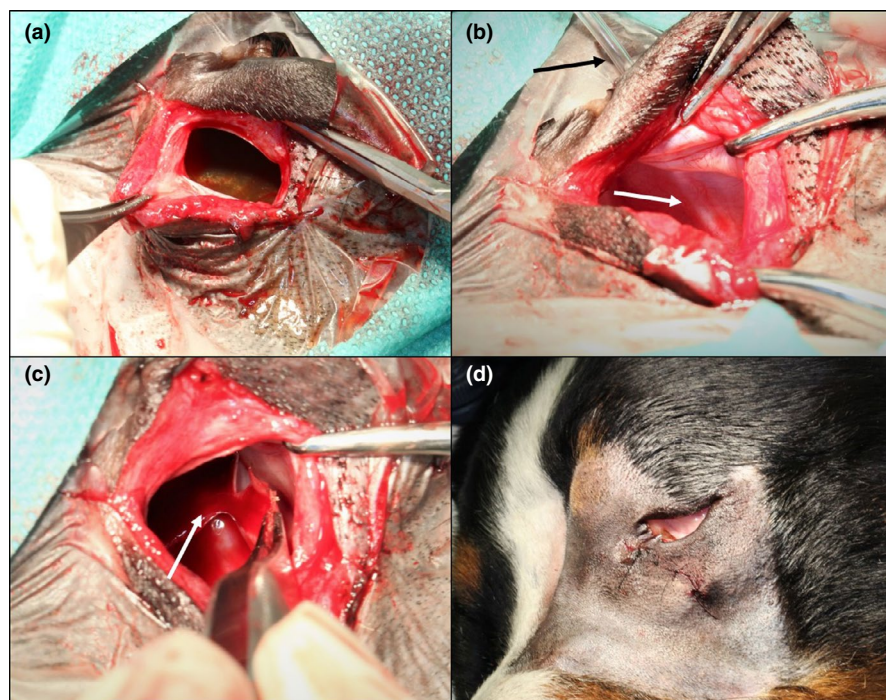


FIGURE 2 Case 3: Cystorhinostomy; (A) view in the cyst which is filled with opaque brownish fluid; (B) the drain (black arrow) in the NLD can be seen outside the cyst (white arrow); (C) resection of the medial cyst wall (arrow); (D) the lateral fibrous and subcutaneous tissues were used to create a firm coverage of the bone defect

The left side was affected in all male dogs; whereas, the female neutered dog presented with the condition on the right side. All dogs in this study showed a unilateral round, indolent, and non-tender cystic process (diameters 1.5–3 cm) medio-ventral to the nasal canthus of one eye in the region of the lacrimal sac and serous discharge in the same eye. Slit-lamp examination did not reveal any other abnormalities. The NLD was obstructed on the affected side before treatment but not on the contralateral side, as determined by the Jones test (cases 1–3).

A large expansive lesion destroying the maxilla and the ethmoid turbinates (partially) could be seen on transverse CT in all four cases. The lesion was filled with dense fluid material that did not demonstrate contrast enhancement (Figure 3). The proximal bony portion of the NLD did not reveal any abnormalities. The NLD could be clearly distinguished from the cystic structure in all cases based on the presence of a thin membrane between the two (Figure 4). Dacryocystorhinography was attempted, however was unsuccessful due to immediate back flow of the contrast medium when injected in the dorsal lacrimal tubule.

Cystic content in all cases was an opaque brownish fluid with some protein, and non-diagnostic cytologic and negative culture findings (cases 1, 2, and 4; see Table 1).

In case 1, the thin medial cystic wall ruptured during the first injection of povidone iodine. After this event, the nose was kept low and the fluid was aspirated immediately. No further complications were observed, and the povidone iodine injection was repeated three months after the recurrence at three weeks. No recurrence was observed after the second sclerotherapy. In case 2, the first injection of povidone iodine was successful with no recurrence within the follow-up period. In case 4, recurrence was noticed three weeks after the injection, and the two dogs (case 3 and 4) underwent cystorhinostomy.

Histopathological examination revealed that the cystic structure was surrounded by an attenuated epithelium consisting of a double layer of cuboidal cells characteristic of excretory ducts of glands. Occasionally, disruption of the epithelial lining (ulceration) was observed with replacement by granulation tissue. The epithelium had

FIGURE 3 Cases 1–4: transverse CT image: a large expansile lesion destroying the maxilla and partially ethmoid turbinates can be seen

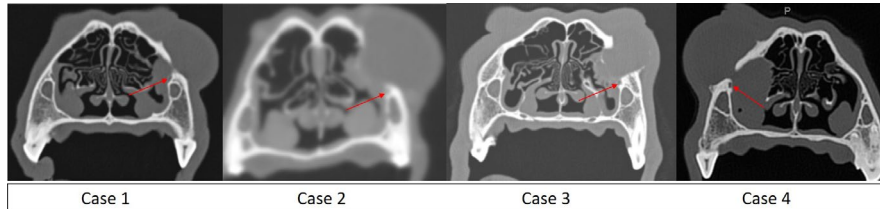
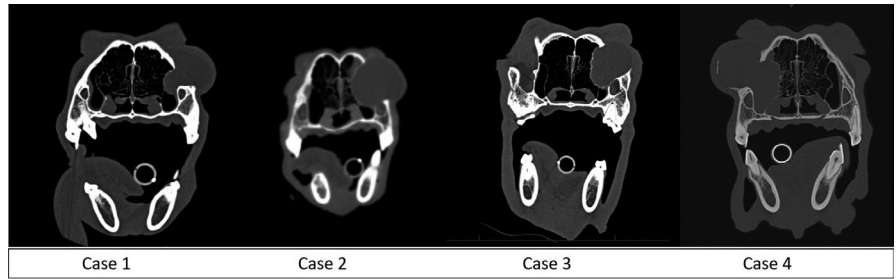


FIGURE 4 Cases 1–4: transverse CT image: the proximal bone part of the naso-lacrimal duct was without abnormalities. The NLD can be clearly distinguished from the cystic structure in all cases by a thin membrane between the two (arrows)

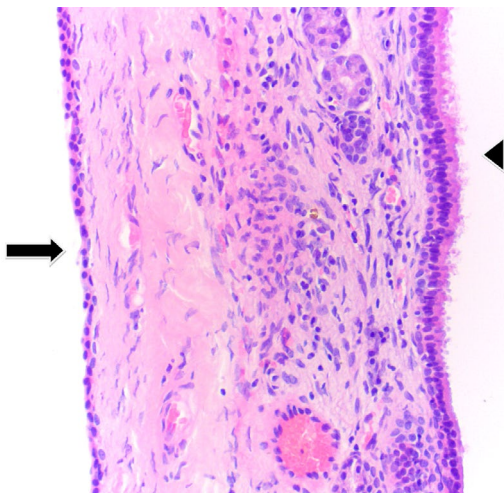


FIGURE 5 Case 3: Histopathological findings of the medial cyst wall: HE staining, 20x, a double layer of cuboidal cells can be seen (arrow), the bone was atrophic, contralateral epithel of the nasal mucosa (arrowhead)

subjacent fibrovascular tissue with embedded apocrine glands, dilated blood vessels, and few macrophages and lymphocytes. The underlying bone was completely atrophied. Normal mucosa was present on the opposite side (Figure 5).

There was no recurrence and very good to excellent cosmetic results were achieved in all animals during the follow-up period (Figure 6). There was no discernible conjunctival irritation or serous discharge. Therefore, the patency of the NLD could be reasonable ascertained.

4 | DISCUSSION

4.1 | Differential diagnosis and etiopathogenesis

The cystic process on the medio-ventral orbital region in the dogs included in this study appeared clinically similar to that of a dacryocystocele (DC) in humans.^{13,14} Dacryocystocele DC is a diffuse enlargement of the lacrimal sac due to narrowing or obstruction of the naso-lacrimal duct. In humans, it is frequently observed in newborns due to congenital lacrimal system stenosis or rarely as acquired DC.^{14–16}

However, in all four animals included in this study, the naso-lacrimal drainage system was intact and did not communicate with the cyst. This was confirmed by cannulation at the time of surgery in cases 3 and 4 [a naso-lacrimal catheter could be placed before surgery and was not visible in the cyst, but was close to the cystic wall (Figure 2B)], CT images, and disappearance of the serous epiphora in all cases after reduction of the cyst. The flow of the NLD was obstructed and compressed from the external aspect only before therapy in all cases. Based on these findings, DC could be ruled out as a differential diagnosis. This is in accordance with the current veterinary literature. Van der Woerd et al¹⁷ identified a cystic dilatation of the naso-lacrimal system distal to the naso-lacrimal sac: focal inflammation of the NLD in three dogs that resulted in obstruction, development of a cystic dilatation in the superior portion of the NLD, and recurrent dacryocystitis. The occurrence of DCs in companion animals is not known because there are no studies on the condition.

The morphological features revealed by histopathological examination supported the diagnosis of a dacryops. A



FIGURE 6 Case 2: clinical presentation post polidocanol; Cases 3 and 4: postsurgical

partially cystic proliferation and a cystic structure surrounded by an attenuated epithelium consisting of a double layer of cuboidal epithelial cells,^{18,19} characteristic of the excretory duct associated with lobules of lacrimal gland tissue clearly suggest cyst formation from a glandular excretory duct and/or glandular tissue.

Dacryops in companion animals have been reported in the conjunctiva and in the ventromedial site of the orbit in dogs^{12,20} and at the third eyelid of cats²¹. In humans, and in the ventromedial site of the orbit in dogs^{12,20,21} dacryops is an uncommon benign cystic lacrimal gland tissue neoplasm. The exact etiology of dacryops remains unknown.²² Cysts of ectopic glands have been reported to persist in some humans¹⁸ and could be considered among animals in the present study as well, as there is no normal lacrimal gland tissue in the ventromedial orbital region. Furthermore, no connection to the lacrimal gland of the third eyelid and the caruncle could be determined in the cases included in this study. The most common location of ectopic lacrimal gland tissue in humans is the epibulbar conjunctiva (62%), and other locations include the orbital (16%), eyelid (11%), intraocular (9%), lacrimal sac (2%), and nasal mucosa (0.6%) sites.²³ One case report on humans stated that the development of an orbital ectopic lacrimal gland cyst is extremely rare.²⁴ One case report on humans stated that the development of an orbital ectopic lacrimal gland cyst is extremely rare.²⁴ It was originally suggested that such cysts form secondary to ductal occlusion and subsequent build-up of secretions; however, this hypothesis has not been supported experimentally. One study discussed dysfunction of the rich neural plexus around the ductules in conjunction with periductular inflammation, which induced scarring. More recently, it has been hypothesized that some variants may form secondary to chronic inflammation of the lacrimal gland, causing a secondary immune response by the conjunctiva-associated lymphoid system with oversecretion of IgA and an osmotic effect resulting in cyst formation.^{25,26}

Green and Zimmermann²⁷ postulated a pathogenesis that small sequestrations could migrate more deeply into the orbital soft tissues when the lacrimal anlage was laid down by evagination of the supratemporal embryonic conjunctival epithelium into the anterior orbital soft tissues. In the case of an orbital cyst without physical association with the NLD in a dog, the authors assumed that the duct epithelium “immigrate” into the adjacent maxillary process and subsequently

developed into an expanding cystic structure.⁶ The findings of the present study provide evidence of a similar mechanism.

4.2 | Diagnostic imaging and therapies

Orbital cysts in humans and companion animals can be identified by sonography,^{12,28} radiography,^{3,6} magnetic resonance imaging,²⁹ and CT imaging,^{4,8–10,14,29,30} and the latter has been preferred in more recent studies. CT features of cysts due to ectopic lacrimal gland tissues have been described as non-aggressive destruction of surrounding osseous structures depending on the location (turbinates, nasal/lacrimal/maxillary bone)^{8,31} as could be seen in all four animals in the present study (Figure 3). The NLD could be clearly distinguished from the cystic structure in all four dogs in this study based on the presence of a thin membrane between them (Figure 4). A normal naso-lacrimal system could be identified which was closely associated, but did not communicate with the cystic mass. This has also been observed in the study by Ota and Pearce⁵ who reported on dacryocystorhinography in three young Labradors as well as in few other case studies conducted on dogs^{1,3,5,6,12} and a horse.⁴

Unfortunately, dacryocystorhinography could not be performed in this study.

Therapies for ventromedial orbital cysts in companion animals reported in other studies include use of a sclerosing agent^{10,11,32} and complete excision.^{1,4,6,9,10,12} DCs in humans, which lie in the ventromedial orbit similar to dacryops in the four dogs included in this study, have been treated by external¹⁴ and endoscopic^{13,16,33} dacryocystorhinostomy.

The sclerosing agent polidocanol 1% has been used with variable outcomes in dogs.^{10,11,32} In this study, polidocanol was applied in cases 1, 2, and 4. Success was achieved in case 2 despite the large size of the cyst; however, polidocanol application resulted in failure in cases 1 and 4. In case 1, the first instillation had to be terminated because the thin medial wall burst during procedure. Even if the second attempt in case 1 was successful and there was no communication in all cases between the cysts and the NLD, polidocanol should be injected carefully considering the risk of complications and/or adverse effects.

Surgical intervention was planned in case 3 as the first choice and in case 4 after failure of the polidocanol injection.

Lateral dacryocystorhinostomy or marsupialisation was chosen to create a firm coverage of the huge bone defect that which would not have been possible by endoscopic intervention. External and endoscopic dacryocystorhinostomy have similar success rates in humans.³⁴

In the case of NLD obstruction alone, canaliculorhinostomy has been reported to be successful in dogs and cats.³⁵ The dogs in the present study showed good functional and cosmetic outcomes.

Limitations of this study include its retrospective nature of the study, small sample size, and the relatively short follow-up period in two cases. Only 1 of the animals included in this study could be examined at our hospital. The others were controlled by a referring veterinarian and/or by photographic documentation. Furthermore, the Jones 1 test or NLD flushing could not be performed after therapy in case 4 because of the challenges in controlling the dog. However, due to patency of the NLD in cases 3 and 4 during surgery and the absence of serous epiphora in all four cases the integrity of the NLD could be strongly supposed.

5 | CONCLUSIONS

In cases of ventromedial orbital cysts, aberrant lacrimal gland, and ductal tissue with secondary development of dacryops should be included in the differential diagnosis. Large cysts near the lacrimal drainage system with extensive bone defects in dogs should be treated by nasal marsupialisation. Evacuation of the cyst and sclerotherapy should be reserved for exceptional cases. Further research is indicated, including immunohistological characterization of the tissues, for clarification of the origin of the cyst in this anatomical region. In addition, prospective studies must be conducted on a larger sample size with surgical intervention.

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