Imaging diagnosis of paranasal sinus mucocele in a Yorkshire Terrier dog

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Abstract: An 8-year-old, neutered male Yorkshire Terrier dog presented with left ventromedial canthus swelling over a one-month period, refractory to pharmacological therapy. There was no history of trauma. On ultrasonography, the lesion had a cystic character filled with anechoic fluid and hyperechoic sedimenting debris. The computed tomography (CT) and CT-dacryocystography showed a cystic lesion protruding from the lacrimal sac fossa and occupying a defect in the orbital plate and an ethmoidal ectoturbinate surrounded by a bony structure with an intact nasolacrimal system. The dog underwent the surgical resection of the cyst and its fluid content was aspirated. Ethmoid mucocele was diagnosed based on the CT, cytologic examination, bacterial culture and histopathologic findings. This case describes the imaging characteristics of an ethmoid mucocele and highlights the importance of CT and CT-dacryocystography in dogs with ventromedial canthus swelling that had poor response to medical treatment.

Keywords: canine; computed tomography; dacryocystography; ethmoid mucocele

Paranasal sinus mucoceles are benign, mucus-containing cystic structures lined by a respiratory epithelium. Anatomically, paranasal sinuses are air-filled cavities lined by a mucoperiosteum that secretes mucus via the sinus ostium to prevent nasal mucosal drying (Capra et al. 2012; Evans and de Lahunta 2013). A paranasal sinus mucocele is thought to result from occlusion of the sinus ostium by inflammation, fibrosis, trauma, abnormal conformation, or a mass; however, the definitive cause remains undetermined (Kawaguchi et al. 2002; Eggesbo 2006). Despite its benign histological nature, it erodes adjacent bony walls and/or expands intracranially (Adamo 2005; Capra et al. 2012; Vandenberghe et al. 2020). Few reports about mucoceles located in paranasal sinuses in dogs and cats have been documented in literature. All of the cases were localised in the frontal sinus (Harvey 1971; Gilson and Stone 1991; Hakanson 1994; L'Eplattenier and

Montavon 1998; Adamo 2005; Sessums and Lane 2008). To the authors' knowledge, there has been no report of ethmoidal mucoceles that could be confused with a nasolacrimal disease. This report presents the case of an ethmoidal mucocele that appeared without a traumatic event, and its computed tomography (CT) and CT-dacryocystography findings that could be helpful to differentiate ethmoidal mucoceles from other diseases that could cause swelling of the ventromedial canthus.

Case description

An 8-year-old neutered male Yorkshire Terrier dog was referred with a one-month history of a slowly growing left periorbital mass (Figure 1A). This lesion had a poor response to the pharmacological therapy, such as prednisolone and antibiotics, while

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Figure 1. Representative extraocular photograph (A) and ultrasonography image [linear transducer, 5–12 MHz (B)] of the ventromedial canthus swelling on the left eye (OS)

Ultrasonography image (B) shows a well-defined, superficial, anechoic, spherical, cystic structure (1.5 cm \times 1.7 cm) with partial posterior acoustic enhancement. A mild amount of hyperechoic sedimentation debris is visible in the cystic structure. This cystic structure compresses and dislocate the OS laterally

the eye discharge was reduced. There was no history of trauma. A physical examination revealed a non-mobile, firm, discrete swelling lesion located in the left ventromedial canthus having a diameter of less than 2 cm. An ophthalmological examination including Schirmer's test showed no remarkable findings. The complete blood count, serum biochemical analysis, and electrolyte examination were all within normal limits.

Ultrasonography (Affiniti 50; Philips Medical Systems, Bothell, WA, USA) with broadband high-resolution linear transducers (5–12 MHz) was performed to evaluate the periorbital swelling. Transverse and longitudinal scans identified a well-defined, superficial, spherical cystic lesion (1.5 cm × 1.7 cm) filled with an anechoic fluid with a partial posterior acoustic enhancement. The hyperechoic sedimentation debris was settling and moving in a gravity-dependent manner in this cystic lesion. The left eyeball was slightly dislocated and compressed by the cystic lesion (Figure 1B).

A CT scan was performed using a 16-row multislice CT scanner (Alexion; Toshiba Medical System, Ohtawara, Japan). The dog was anaesthetised using propofol (5 mg/kg, i.v., Provive; Myungmoon Pharm Co., Seoul, Republic of Korea) for induction and isoflurane (2–3% inspired volume, Ifran; Hana Pharm Co., Seoul, Republic of Korea) and oxygen (1.0–1.1 l/min) for maintenance through an endotracheal tube. The patient was placed in sternal recumbency on the CT table. The scanning parameters were a contiguous slice thickness

of 2 mm, 150 mA, and 120 kV. The CT data were reconstructed with four cross-sectional images (transverse, sagittal, dorsal plane, and oblique) and three-dimensional images. The three-dimensional images were obtained from the CT images with a 1.0 mm slice thickness and 0.8 mm slice intervals. Contrast studies were performed after the intravenous administration of Iohexol (600 mg iodine/kg, Omnipaque 300; GE Healthcare, Cork, Ireland) injected for 20 s using an autoinjector (Medrad Vistron CT Injector System; Medrad, Indianola, USA). The arterial and delayed phases were acquired 20 s and 90 s after injection, respectively. The non-contrast CT revealed an oval shaped fluid-attenuating lesion (mean, 18 HU) sized $1.9 \text{ cm} \times 1.7 \text{ cm} \times 2.0 \text{ cm}$. This cystic structure was protruding from the left lacrimal sac fossa and occupying a defect in the orbital plate and ethmoidal ectoturbinate in front of the infraorbital margin. The stalk of this cystic mass was in the orbital plate of the ethmoid bone. There was a disruption in the maxillary bone and this cystic mass was partially surrounded by a bony structure protruding from the lacrimal sac fossa with a bone dehiscence. On the post-contrast CT, a peripheral ring enhancement, the central attenuation remained unchanged indicative of compartmentalised fluid, was identified (Figure 2).

Consequently, dacryocystography using CT was performed to determine whether the nasolacrimal duct was involved. In the same sternal recumbency position, the dorsal punctum of the left eye was gently cannulated with a stylet-removed 24-gage

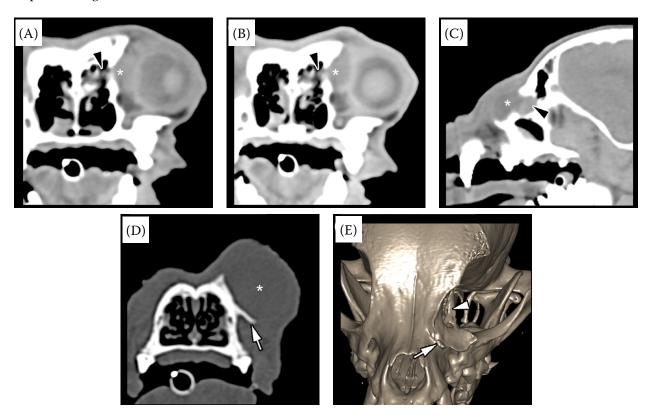
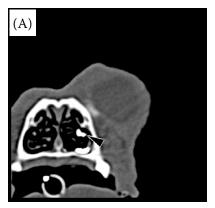


Figure 2. CT images of the head

Unenhanced (A), contrast-enhanced (B) transverse and reconstructed oblique plane (C) CT in a soft tissue window setting (window width 450 HU, window level 40 HU) shows a non-contrast enhanced cystic lesion (18 HU, asterisk) with a ring enhancement in the caudal part of the left maxillary bone in front of the infraorbital margin. This cystic mass communicates with the nasal concha (black arrowheads) via a bone defect in the orbital plate. Transverse plane (D) with a bone window setting (window width 2 000 HU, window level 600 HU) and a 3D-CT (E) show the new bone formation (arrows) surrounding the cystic mass (asterisk) with the bone erosion of the adjacent maxilla (white arrowhead)

catheter and approximately 2 ml of Iohexol diluted with eyewash was injected. The contrast medium was injected without resistance. The CT scans were performed after the contrast medium was noted

to exit from the nares. The CT-dacryocystography showed an intact nasolacrimal drainage and patency without disruption or dilation of the nasolacrimal duct (Figure 3).



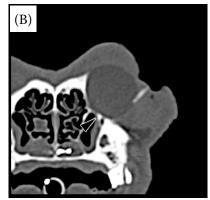




Figure 3. CT-dacryocystography with a bone window setting (window width 2 000 HU, window level 600 HU) At the level of the nasolacrimal duct opening (A) and nasolacrimal canal (B), the patency of the lacrimal drainage is confirmed by detection of the contrast medium in the nasal meatus (arrowhead). The reconstructed dorsal plane (C) shows the intact left nasolacrimal duct and canal (arrows). The cystic lesion located in the left maxilla is not related to the nasolacrimal duct

A surgical exploration of the cyst was performed after the CT scans. Before the surgical resection, a small amount of the cystic fluid was aspirated for the laboratory examination and a colourless, viscous fluid was collected. Upon surgery, the cystic lesion adhered firmly to the peripheral tissue, especially to the eroded maxillary bone. The cystic structure, peripheral bone, and soft tissue were removed. The aerobic and anaerobic bacterial cultures of the fluid were negative. The cytologic examination of the cystic fluid primarily yielded foamy macrophages and few neutrophils, consistent with mild nonspecific, chronic inflammation. There was no evidence of a significant infection

or any malignant cells. The histopathological examination of the pericystic tissue was consistent with a chronic inflammation with granulation and haemosiderophages. An ethmoidal mucocele was diagnosed on the basis of the imaging, histopathological, and bacteriological findings.

Follow-ups were performed every month and there was no recurrence. During a follow-up 9 months post operation, swelling on the left medial frontal region above the eyeball was palpated. This mass showed similar results to the previous examination, in terms of the ultrasound, CT (Figure 4), cytology of the cystic fluid, and bacterial culture, hence a frontal sinus mucocele was diagnosed.

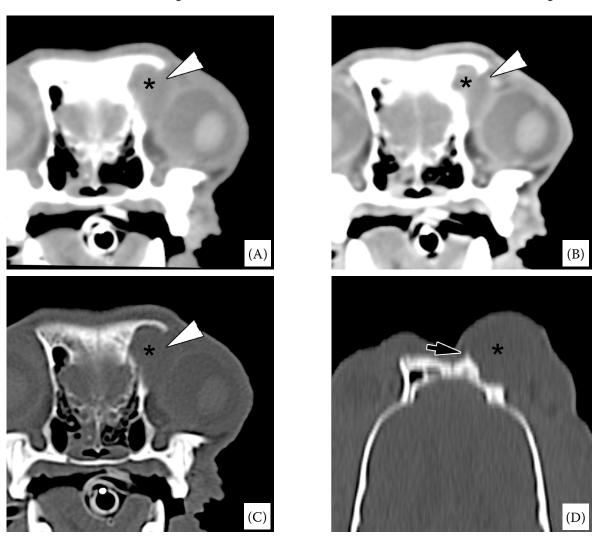


Figure 4. CT images of the head

Unenhanced (A) and contrast-enhanced transverse (B) CT with a soft tissue window setting (window width 450 HU, window level 40 HU) show a non-contrast enhanced cystic lesion (35 HU, asterisk) with a ring enhancement in the left frontal sinus. The wall of the frontal sinus has a defect (arrowheads). The transverse (C) and reconstructed dorsal plane (D) CT with a bone window setting (window width 2 000 HU, window level 600 HU) show the new bone formation (arrow) surrounding the cystic lesion (asterisk)

DISCUSSION AND CONCLUSIONS

In veterinary medicine, the list of differential diagnoses for swelling of the ventromedial canthus include dacryocystitis, dacryops, cystic dilatation of the canaliculi, cysts of the orbital adnexal tissue, abscesses, granulomas, congenital dermoid cysts, and neoplasms (Cullen and Grahn 2003). Because of the anatomical proximity of the nasolacrimal duct and the medial canthus, the differentiation, for appropriate treatment, is focused on assessing the involvement of the nasolacrimal duct. In this case, a patient presented with left ventromedial canthus swelling, refractory to the pharmacological therapy, and there was an eye discharge prior to the pharmacological therapy. The medical history of the patient suggested the need to evaluate the nasolacrimal system, and additional diagnostic examinations were required.

Diagnostic imaging, such as a conventional dacryocystography using a radiograph, CT, or magnetic resonance imaging (MRI), have been used for evaluating nasolacrimal diseases (Nykamp et al. 2004; Raslan et al. 2019). MRI has the advantages of showing a higher soft tissue contrast resolution that allows for the better assessment of the periorbital soft tissues and is free from ionising radiation on the lens. However, it is limited to assessing the bony nasolacrimal canal while a CT has the best modality for delineating the nasolacrimal drainage apparatus within a short scanning time (Raslan et al. 2019). In addition, the CT has the advantage of being able to perform a CT-dacryocystography following the examination. For these reasons, the CT and CT-dacryocystography were performed to identify the nasolacrimal system.

In previous studies of veterinary CT-dacryocystography, Nykamp et al. (2004) suggested four considerations when evaluating a medial canthus mass: (1) location, (2) opacity and margin, (3) the presence of bone remodelling or lysis, (4) the presence of disease in the adjacent nasal cavity. According to these considerations, Nykamp et al. (2004) suggested several differential diagnoses of dacryops, mucoceles, abscesses, neoplasia, or infections of the nasolacrimal apparatus. Also, this previous study emphasised the need for CT-dacryocystography when the lesion is not located over the nasolacrimal apparatus to assess the secondary involvement of the nasolacrimal duct (Nykamp et al. 2004). According to the previous differentials, the cystic

lesion of this patient was nearly similar to dacryops or a mucocele, in that the lesion had smooth margins and was a cyst-like structure, but considering that the cystic lesion was not filled with contrast medium in the CT-dacryocystography, the dacryops and nasolacrimal mucocele were excluded. In addition, the presence of osteolysis in this case obscures this classification by showing the possibility of a tumour and infections. Thus, in addition to the prior differential, when the cystic lesion is in the lacrimal sac fossa with osteolysis on the CT, an ethmoidal mucocele should also be considered, and the patency of the nasolacrimal system should be evaluated using CT-dacryocystography.

In veterinary medicine, there have been few reports of diagnostic imaging findings of a paranasal sinus mucocele, as these are mainly about the frontal mucocele with an intracranial extension or traumainduced (Adamo 2005; Sessums and Lane 2008; Raul et al. 2018; Vandenberghe et al. 2020). On the CT, the paranasal mucocele is shown as a homogenous, hypoattenuating area with a ring enhancement (Raul et al. 2018) like that in our case also. On the MRI, a paranasal sinus mucocele is shown as heterogeneously hypointense on the T1-weighted and hyperintense on the T2-weighted images, but this can vary depending on the component of the mucocele (Sessums and Lane 2008; Vandenberghe et al. 2020). In addition to the above imaging findings, in the case of an intracranial extension, a well-demarcated heterogeneous lesion involving all frontal sinuses extends into the intracranial space via the focally-thinned frontal bone (Adamo 2005; Sessums and Lane 2008; Vandenberghe et al. 2020). In a trauma-induced paranasal sinus mucocele, destruction of the cranium with multiple irregular osteolysis may be found (Raul et al. 2018). The reason for the extension of the paranasal sinus mucocele outside the sinus wall is due to the pressure-related bone erosion. When the mucus accumulates in the limited space of the sinus, increasing the pressure can cause atrophy or erosion of the bone, and expand to the path of least resistance, such as the orbit, adjacent sinuses, nasal cavity, cranium, or through the skin (Raul et al. 2018). Similar to the previous report, the patient had a well-demarcated, hypoattenuating mass with contour enhancement disrupting the maxillary bone with some bony structure protruding from the lacrimal sac fossa. These findings may be helpful to differentiate a paranasal sinus mucocele from other diseases such as sinusitis or sinus tumours.

The presence of an ethmoid sinus is disputable. Both the presence and absence in dogs, are described in the literature (Kealy et al. 2010; Evans and de Lahunta 2013). In this case, we used the term ethmoid mucocele in that the mucocele occupied a defect in the orbital plate and ectoturbinate of the ethmoid bone. In previous paranasal sinus mucocele reports in dogs and cats, all mucoceles originated from the frontal sinus, in contrast with the ethmoidal ectoturbinate in this case (Harvey 1971; Gilson and Stone 1991; Hakanson 1994; L'Eplattenier and Montavon 1998; Adamo 2005; Sessums and Lane 2008). The frontal sinuses are the largest while ethmoid sinuses when are described as small (Kealy et al. 2010). This anatomical size difference was thought to have influenced the distribution of the previously reported paranasal sinus mucocele locations in dogs.

In this case, a frontal sinus mucocele on the same side as the prior ethmoid mucocele was diagnosed 9 months after surgery. Anatomically, frontal sinuses are partially divided into the rostral, lateral, medial, and caudal compartments and drain separately into the nasal cavity via the ostia formed by ethmoidal conchae. Among these compartments, the rostral part of the frontal sinus runs forward, adjacent to the orbital plate of the ethmoid bone (Kealy et al. 2010; Evans and de Lahunta 2013). Thus, the anatomical proximity of the frontal sinus and ethmoidal ectoturbinate may have caused the recurrence in this case. In humans, recurrence of paranasal sinus mucoceles have been described. In dogs and cats, however, no recurrence of paranasal sinus mucoceles have been described, and only few reports about paranasal sinus mucoceles exist (Picavet and Jorissen 2005; Mayne et al. 2012). According to previous studies, humans have a high recurrence rate (23.5%) of primary or second paranasal mucoceles (Picavet and Jorissen 2005; Mayne et al. 2012). The incomplete marsupialisation of the primary lesion and unfavourable scarring of a sinus ostium or free mucosal edge have been discussed as the reasons for the recurrence and the presence of acute infections, multiple mucoceles, and a significant extension outside the sinus wall are risk factors for the recurrence (Picavet and Jorissen 2005; Mayne et al. 2012; Bleier et al. 2018). Based on these facts, it was thought that the occurrence of a secondary mucocele in this case may be caused by the scarring of the sinus ostium, or free mucosal edge, due to the surgical procedure, leading to mucus accumulation in the frontal sinus that is adjacent to the ethmoidal ectoturbinate. In addition, this case partially coincided with a significant extension outside the sinus wall, a risk factor for the recurrence of a paranasal sinus mucocele.

The limitation of the present study was that the contrast images of the entire nasolacrimal apparatus were not obtained. This was because a CT scan was not performed while the contrast medium remained in the nasolacrimal duct, like previous CT-dacryocystography studies (Nykamp et al. 2004; Rached et al. 2011). To obtain the contrast images of the entire nasolacrimal system, a CT scan should be performed as soon as the contrast medium is detected in the nares. However, this method increases the radiation exposure for the practitioner, as the practitioner should be in the CT room during the manual contrast medium injection. It may also increase the radiation exposure for the patient as a dynamic CT scan of the nares should be performed. Though the contrast images of the entire nasolacrimal apparatus were not acquired, this did not affect the diagnosis.

In addition, because the contrast medium remains in the lesion or obstruction site in the case of a nasolacrimal disease, the CT-dacryocystography in this case was thought to be enough to identifying the patency of the nasolacrimal system to rule out an obstruction.

In summary, this report described the imaging findings of an ethmoid mucocele in a dog without trauma, and this is the first case reported of an ethmoid mucocele. CT and CT-dacryocystography should be performed in dogs with a ventral inner canthus mass that has a poor response to a pharmacological treatment to differentiate from a nasolacrimal disease. If the CT reveals a well-demarcated cystic lesion with a ring enhancement in the lacrimal sac fossa with adjacent osteolysis, an ethmoid mucocele should be considered once the patency of the nasolacrimal system has also been established.

Conflict of interest

The authors declare no conflict of interest.

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