Differential diagnoses, investigation and management of a periocular swelling close to the nasolacrimal duct in a horse – A case report of Dacryops

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Running Title: Dacryops in a horse

Abstract:

Objective: To describe the differential diagnoses, investigation and management of a periocular swelling close to the nasolacrimal duct in a horse that was consistent with a nasolacrimal duct dacryops (lacrimal cyst).

Animal studied: A 16 year old gelding, Connemara cross.

Procedures: The horse presented with a history of a periocular swelling rostro-ventro-medial to the right eye that had been sampled by the referring veterinary surgeon. A cystic lesion was diagnosed following standing computed tomography. Surgical removal of the cystic lesion was performed and the tissue was submitted for histopathologic and immunohistochemical examination.

Results: Surgical removal of the cyst was curative and there was no recurrence of clinical signs 7 months later. There was a small amount of mineralized material in the center of the cyst. Histopathologic and immunohistochemical examination confirmed a nasolacrimal duct cyst.

Conclusion: Dacryops can form in horses as well as other species and appears to have a favorable outcome if surgically removed.

Key Words: nasolacrimal system, cyst, equine, dacryops, computed tomography, histopathology

Word count: 154

Introduction:

Dacryops, or lacrimal cysts are fluid filled distensions of the lacrimal system and have been reported in the dog (1), red-eared slider (2) and people (3). It has also been described in different locations such as the lacrimal gland (4), ventromedial conjunctiva(5), and nasolacrimal duct itself(1). Histopathological examination of the tissue reveals an epithelial lining of stratified squamous origin(6). Mineral deposits have been reported within the nasolacrimal system in dogs(6) and horses(7). The canine deposit was reported to be calcium carbonate composition in the dog(6) and was a mixture of, mainly carbonate hydroxyapatite, and a small amount of sodium chloride halite in the horse(7). To the authors' knowledge this is the first reported case of dacryops in the nasolacrimal system of a horse.

Case report:

A 16-year-old Connemara cross gelding presented to the Royal Veterinary College, Equine Referral Hospital with a two month history of swelling located rostro-ventro-medial to the right eye. Fluid-like material had been aspirated by the referring veterinarian two months prior to referral. Cytologic evaluation was consistent with neutrophilic inflammation.

A complete ophthalmic examination revealed that both eyes were comfortable and visual. The menace response was present in both eyes and all reflexes were present. Schirmer Tear Test – 1 readings were 18mm/min in each eye. All ocular media were clear and funduscopy was consistent with a healthy equine fundus. Intraocular pressures were 25mmHg in both eyes (Tonovet® Tiolat, Helsinki, Finland). Fluorescein (Minims, Bausch & Lomb House, surrey, UK) was negative in both eyes and the Jones Test was positive bilaterally. The nasolacrimal ducts were irrigated in a

retrograde direction and with ease. Following aspiration by the referring veterinarian, the swelling immediately resolved and was not evident of referral examination. At this time differential diagnoses included: Nasolacrimal duct cyst (congenital or acquired), cyst unrelated to the nasolacrimal duct, foreign body granuloma, suture exostosis, and neoplasia. Potential further diagnostics included plain radiography, plain computed tomography (CT) or dacryorhinocystography with either radiography or CT. After discussion with the owner, further diagnostic imaging was performed to assess for any structural changes that may have been present in light of the apparent resolution of the swelling. CT was the imaging modality of choice due to a superior anatomical understanding of where any pathology lies and avoidance of superimposition of structures. At this stage contrast dacryorhinocystography was not considered due to normal irrigation of the nasolarimal duct and lack of evidence of swelling.

The patient was sedated using intravenous (IV) acepromazine maleate (Novartis, Surrey, UK, 0.05mg/kg), detomidine hydrochloride (Domosedan, Orion Corporation, Espoo, Finland, 10µg/kg) and butorphanol tartrate (Zoetis, London, UK, 0.1mg/kg). A standing computed tomography (CT) scan was performed with a 16 slice scanner (GE LightSpeed Pro 16, GE Medical Systems, Berkshire, UK) using 1.25mm thick slices and a 1.25mm interslice gap, 120kVp and 200mA with a tube rotation time of 0.5 seconds. CT images of the head were reviewed in a bone and soft tissue window and revealed a single focal 5mm spherical area of gas attenuation within soft tissue surrounded by a thin bone attenuating rim overlying the lateral aspect of the right lacrimal bone immediately rostral to the orbit and adjacent to the right nasolacrimal duct. On a multiplanar reconstruction (MPR) of the CT images there was a suggestion of a complete defect within the lacrimal bone, which could not be clearly appreciated

on transverse images. Concurrent dental disease was identified, with the crown and rostral roots of the 106 tooth being absent and there was a single, clearly marginated vertically orientated hypoattenuating line within the caudal half of the remaining portion of the 106. There was a single roughly diagonal hypoattenuating line within the right maxillary bone overlying the buccal rostral aspect of the 106 alveolus, and an apparent communication from the oral cavity to the alveolar bone in the rostral aspect of the socket of 106 (Figure 1).

Due to the mass no longer being present on examination, and the CT findings being inconclusive, the horse was discharged from the hospital and no medication was prescribed, with instructions for the owner to arrange a reexamination if clinical signs recurred.

The patient re-presented eight months later with a recurrence of the swelling, rostro-ventro-medial to the right eye (Figure 2).

The second ophthalmic examination revealed that both eyes were visual and comfortable and that all reflexes and responses were present and as expected. There was a firm swelling, approximately 4 cm in diameter, situated in the medial canthus of the right eye. The location of the swelling was in close apposition to the right nasolacrimal system. All other aspects of the ophthalmic examination were unremarkable. The Jones test was positive in the left eye and negative in the right eye. Nasolacrimal irrigation was not possible on the right from either direction and no change was appreciated in swelling size and was possible with ease on the left side.

The patient was sedated using the same protocol as stated above, for repeat standing CT. This revealed that immediately rostral to the right eye there was a focal uniformly

soft-tissue attenuating swelling (mean attenuation of 23HU) overlying a smoothly marginated and focally thickened contour of the right lacrimal bone. Further rostral to this area the soft tissue swelling enlarged and there was a concave appearance of the lacrimal bone. The right nasolacrimal duct was normal in size, but had a thickened wall adjacent to the endosteal surface of the lacrimal bone and appeared to communicate with the swelling at a level close to the medial canthus of the eye (Figure 3). These CT findings were consistent with a cyst.

Possible treatment options for this case included: medical management with a sclerosing agent such as polidocanol or surgical removal. Options were discussed with the owner and surgery was chosen.

Intravenous premedication consisted of 0.04 mg/kg of acepromazine IV, xylazine (Chanazine; Chanelle Animal Health, UK) 0.5 mg/kg IV and flunixin meglumine IV (Finadyne, MSD Milton Keynes, UK 2.2mg/kg IV); followed by IV induction with ketamine (Ketaset, Zoetis, UK) at 2.2 mg/kg in combination with midazolam (Hypnovel; Roche, UK) at 0.04 mg/kg mixed in the same syringe. A thiopentone bolus (Link Pharmaceuticals Ltd, UK) of 1 mg/kg IV was required to achieve endotracheal intubation. Once the airway was secured, isoflurane (IsoFlo®; Abbott Animal Health, UK) vaporized in medical air and oxygen mixture (fraction inspired of oxygen of 60%) was administered via a large animal circle system (Mallard Medical Inc., CA, USA). The patient was positioned in left lateral recumbency.

Prophylactic antibiosis consisted on IV penicillin (Crystapen, MSD Milton Keynes, UK) and gentamicin (Dechra, Shrewsbury, UK) before the surgery.

The area around the eye and the nares were prepared with 1:10 dilution of povidone iodine solution, and anterograde cannulation of the nasolacrimal duct was attempted with USP 0 ethilon (Ethicon, Norderstedt, Germany), however this was not possible. An incision was made with a no.11 scalpel blade over the cyst, and scissors were used to bluntly dissect the tissue around the cyst (Figure 4). During the blunt dissection, the cyst ruptured at the site next to the lacrimal bone. Some of the fluid that released was sampled with a charcoal swab, and another sample of the fluid was placed in an EDTA tube. The cyst capsule was removed and placed into 10% neutral-buffered formalin for histopathologic examination.

The nasal opening of the nasolacrimal duct was cannulated with a size 6 urinary catheter. Sterile sodium chloride (Vetevex, Shrewsbury, UK) was irrigated in a retrograde direction, and was seen to exit the upper and lower puncti of the right eye. There was no exit of fluid at the site of the cyst, indicating the nasolacrimal duct was intact. The nasolacrimal duct and the cyst were closely associated, however appeared not to be directly communicating.

The subcutaneous layer was closed with USP 2-0 polyglactin 910 (Vicryl, Ethicon, Norderstedt, Germany) in a simple continuous pattern.

USP 2-0 polyglactin 910 (Vicryl, Ethicon, Norderstedt, Germany) was used to place intradermal sutures, and USP 2-0 ethilon (Ethicon, Norderstedt, Germany) was used to suture the skin with a simple interrupted pattern.

The day following surgery, antibiotic treatment was changed to oral trimethoprim sulphonamides (Noradine, Norbrooks, Northhampton, UK, twice daily for 10 days) and nonsteroidal anti-inflammatory treatment changed to phenylbutazone (Equipalazone, Dechara, Shrewsbury, UK, twice daily for 2 days then once daily for 5 days) orally.

The patient was discharged the day after surgery. The wound was assessed by the referring veterinary surgeon and the sutures were removed without complication two weeks postoperatively. Seven months post operatively the owner reported that there was no recurrence of the swelling.

Histopathologic examination of the resected tissue revealed dense collagenous and vascular connective tissue with a partially collapsed, central cystic space, lined by a single layer of attenuated epithelium (Figure 5). Within the surrounding fibrous connective tissue there were scattered smaller cystic spaces often lacking an epithelial lining and containing scant mucinous material. These were surrounded by numerous clear acicular (cholesterol) clefts, areas of hemorrhage and aggregates of macrophages containing hemosiderin. There were multifocal small aggregates of lymphocytes and plasma cells within the tissue. The structure was diagnosed as a focal lacrimal duct cyst with mild lymphoplasmacytic cellulitis and acute and chronic hemorrhage.

Immunohistochemical staining for pancytokeratin AE1/3 (Dako, Ely, United Kingdom) and alpha smooth muscle actin (α-SMA, Leica, Milton Keynes, United Kingdom) was performed using a BondMax Autostainer (Leica). Both assays used a standard Leica protocol with heat-induced antigen retrieval and the Bond Polymer Refine Detection system (Leica) and with pancytokeratin at 1:100 and α-SMA at 1:250 concentration. The cyst lining cells were strongly positive for pancytokeratin, confirming an epithelial origin, and a subset of spindloid cells, immediately deep to the epithelium were strongly positive for alpha smooth muscle actin, confirming a myoepithelial origin (Figures 6 and 7). No bacteria were isolated after 48 hours incubation, aerobically or anaerobically or after selective culture.

Analysis of the mineral material found within the cystic lesion was not performed.

Discussion:

The presentation of this case is similar to that of previously reported in dogs (1,6). The etiology of dacryops still remains unknown and some authors have described that it could be congenital(8), related to trauma(5), or secondary to chronic dacryocystitis(9). This horse was not young, and given the progression during the 8-month period between evaluations, the authors theorize that the aetiology was unlikely congenital, and more likely to be either traumatic or inflammatory in origin, e.g. recurrent subclinical dacryocystitis. In the human literature it is theorized that periductal inflammation or trauma can stimulate hypersecretion (10). This can lead to cyst formation due to weakening of the ductal and subsequent dilatation (10). Another theory in dogs is ectopic lacrimal tissue causing or predisposing an individual to dacryops (11). In this case an acquired nasolacrimal cyst was felt to be the most likely diagnosis in light of the clinical presentation, immunohistochemical examination and that no lacrimal tissue was demonstrated on histopathological examination. At the time of surgery it appeared that the nasolacrimal duct remained intact, however it is possible if there was an area of stenosis, fibrosis or narrowing of the cyst opening, which could have prevented the fluid from leaking.

Smooth muscle actin immunostaining has been performed in dogs to confirm the diagnosis of dacryops (1). This procedure stains myoepithelial cells in the lining of the cyst. In this case, immunostaining was performed and supported the clinical diagnosis for a dacryops.

The lacrimal system has been previously evaluated with computed tomographic dacryocystography(12,13). In horses, magnetic resonance imaging has also been used to identify the cross sectional anatomy of the lacrimal system (14). Computed

Tomography dacryorhinocystography was not performed due to the technical difficulties experienced specific to this individual patient. In this horse, even non-contrast enhanced CT acquisition was very challenging due to an uncooperative temperament. Despite the appropriate administration of sedatives, it was felt that for safety of staff and equipment that dacryorhinocystography was not an appropriate option. The authors do however recognize that CT dacryorhinocystography would have been beneficial to the optimal diagnostic investigation of this case and that in the general equine population this is a relatively straightforward diagnostic technique. In this case it is likely that CT dacryorhinocystography would have demonstrated communication from the nasolacrimal duct with the lesion preoperatively, and it is a vital tool in the investigation of periocular swelling. Despite this limitation, non-contrast enhanced CT provided adequate information regarding the anatomical location and attenuation characteristics to suggest that a cystic lesion was the cause.

The first computed tomographic examination of this patient was inconclusive because of recent aspiration of cyst contents, reducing visibility and anatomic identification of the cyst directly. It is recognized that CT dacryorhinocystography would have been helpful, however as described above due to patient specific factors this was not possible. It is likely that the second CT was more diagnostic because the cystic cavity had refilled with fluid, making more detailed anatomic localization of the lesion possible. The described CT abnormalities regarding the 106 tooth root (thought to be most likely a primary dental disease) were felt to be an unrelated finding.

Surgical removal of these lesions is usually curative(1,4,5,6,9) and this appeared to be the same in this case. A longer follow up period and a larger sample size would be needed to demonstrate this definitively. Surgical removal is vital as these cysts are

unlikely to spontaneously regress. Without surgery, these cysts lead to eyelid conformational abnormalities such as entropion, and occlusion of the nasolacrimal duct and resulting chronic epiphora (10).

Mineral analysis of the deposits would have been interesting to report if they were similar to the previously reported equine and canine cases. It is unlikely, however, that this would have changed the outcome for this patient.

Conclusion:

Cystic lesions of the lacrimal system can occur in different species and to the authors' knowledge this is the first reported case of dacryops in a horse. It is important to consider this as a differential diagnosis in periocular and nasolacrimal swellings in order to give appropriate guidance and prognosis to referring veterinary surgeons and the owners. Advanced diagnostic imaging and histopathological examination is paramount to a rapid and correct diagnosis.

Figure 1: Transvers CT image showing the first CT performed, revealing a soft tissue attenuating region lateral to the right lacrimal bone (arrow) with a single gas bubble within this lesion.

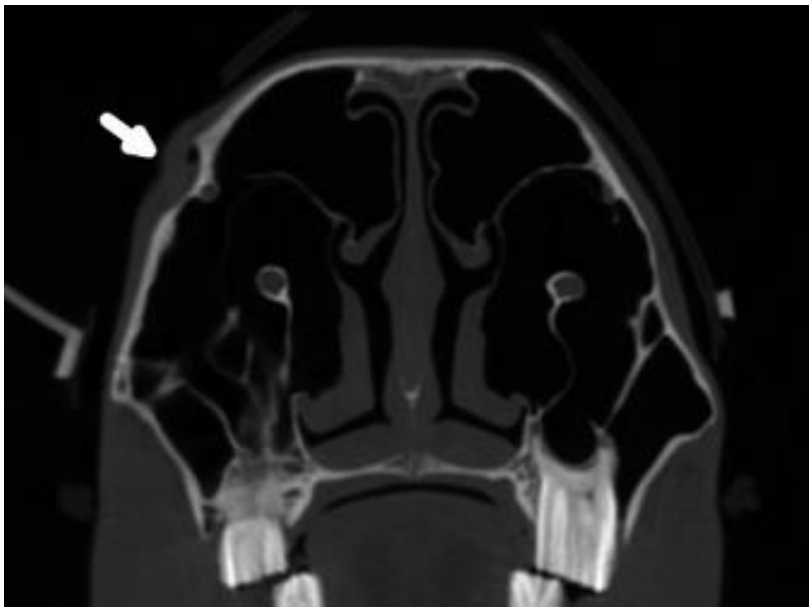


Figure 2: Photograph of the horse preoperatively showing the mass at the rostroventromedial aspect of the right eye.

Figure 3: Transverse CT image showing the cystic lesion (arrow) lateral to the right lacrimal bone, which was enlarged as compared to the previous CT examination.

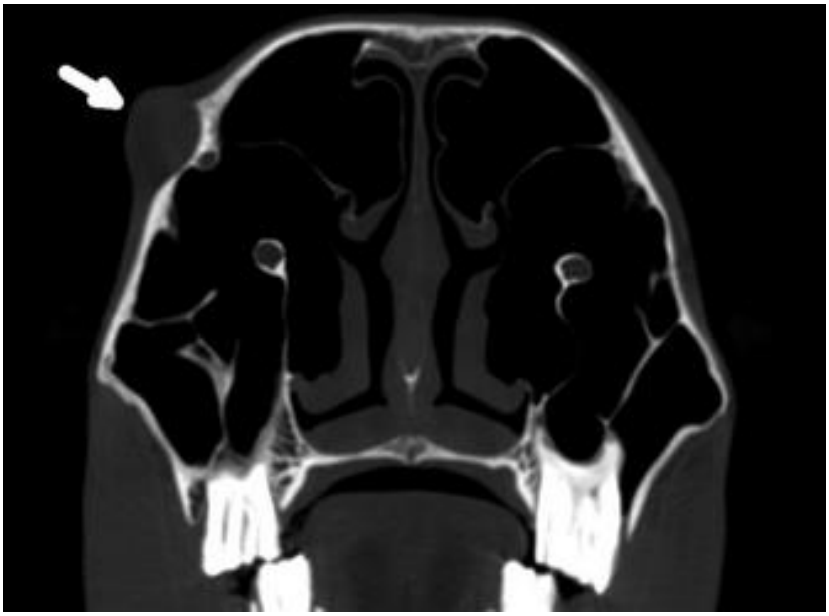


Figure 4: Intraoperative photograph of the cyst (arrow).

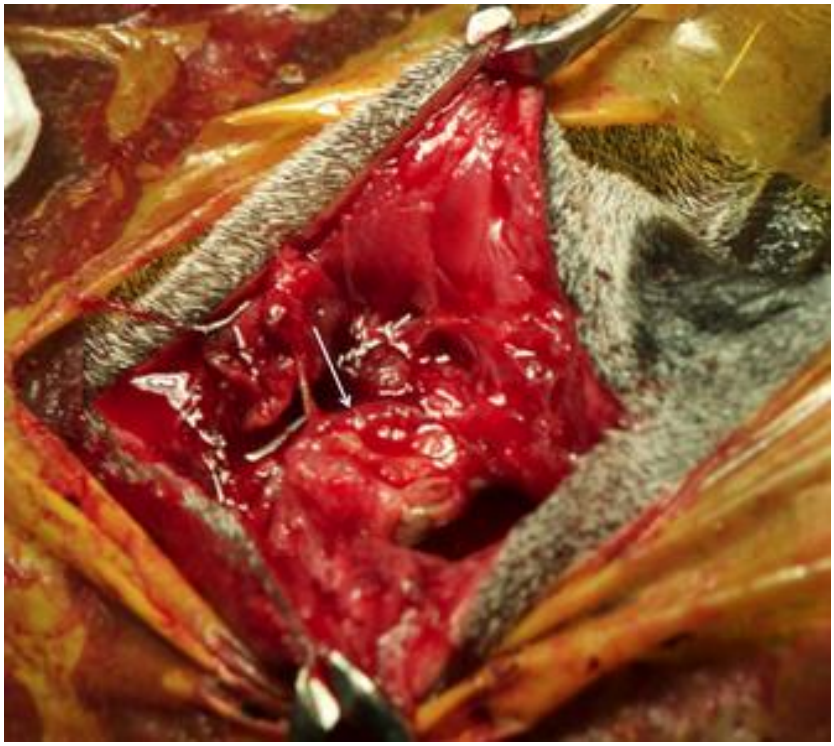


Figure 5: Light microscopy image showing a thick-walled cystic structure, with an attenuated epithelial lining, and multifocal haemorrhage and haemosiderin. Magnification is ×100, bar = 250 micrometers.

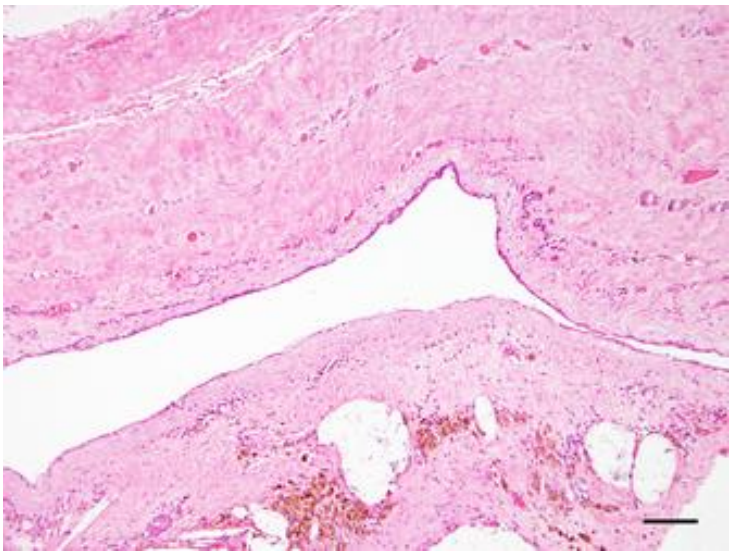


Figure 6: Immunohistochemical staining with pancytokeratin AE1/3 showing the epithelial lining of the cyst. Magnification is ×400, bar = 50 micrometers.

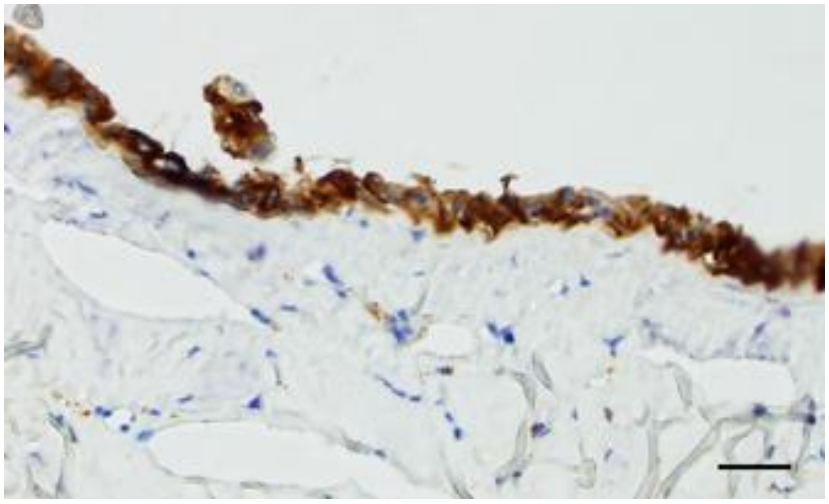
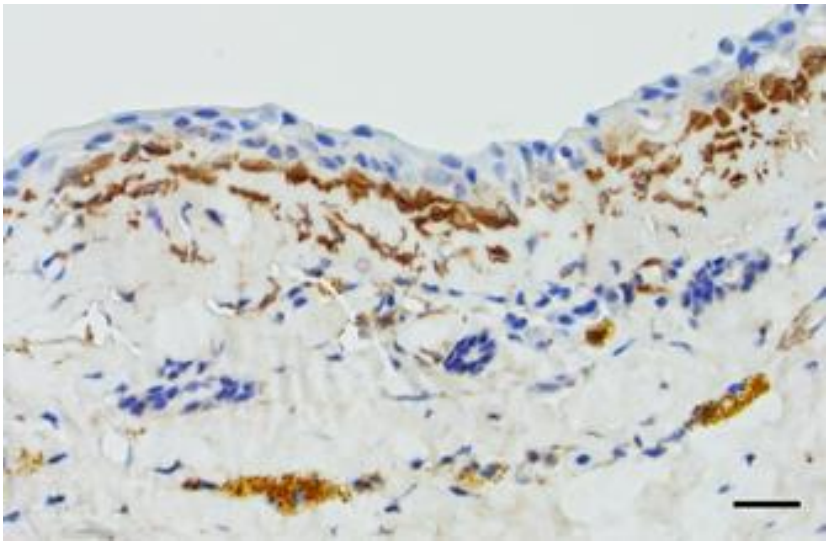


Figure 7: Immunohistochemical staining with alpha smooth muscle actin showing the myoepithelial cells. Magnification is ×400, bar = 50 micrometers.



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