




Case Report

A retrobulbar dermoid cyst with involvement of the sinus in an 18-year old pony

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Summary

This case report describes a retrobulbar dermoid cyst in an 18-year-old pony with an invasive character into the frontal sinus. CT imaging was important for the appropriate management of this case and the definitive diagnosis was obtained from histopathology. Surgical excision was performed successfully without recurrence 1-year post-operatively. The pony returned to its intended use and previous performance level, although complaints of mild symptoms of headshaking were noted in bright light only.

Introduction

Dermoid cysts are uncommon in horses and seen most commonly in younger horses, ranging from 6 months to 9 years of age (Muñoz *et al.* 2007; Knottenbelt *et al.* 2015). Locations for dermoid cysts to occur are the base of the ear (dentigerous cysts), false nostril, eyelid margin, conjunctiva and mandibles (Hillyer *et al.* 2003; Bienert-Zeit *et al.* 2011; Greenberg *et al.* 2012; Knottenbelt *et al.* 2015). Other locations reported for dermoid cysts are the dorsal midline, the ventral thorax and distal limbs (Hillyer *et al.* 2003; Knottenbelt *et al.* 2015). A dermoid cyst in a 4-year-old Andalusian horse developed in the retrobulbar area causing exophthalmos (Muñoz *et al.* 2007).

Histopathologically dermoid cysts are characterised by their adnexal structures, with a central cavity containing keratin and hair shafts (Knottenbelt *et al.* 2015). A granulomatous inflammation can be seen in the surrounding soft tissues where the keratin has escaped from the cyst (Knottenbelt *et al.* 2015).

This case report describes a retrobulbar dermoid cyst in an 18-year-old pony with an invasive character into the frontal sinus. To the authors knowledge, a dermoid cyst in a pony of this advanced age and the invasive character into the sinus, diagnosed by CT-scan, has not been described before.

Case history

An 18-year-old NRPS pony (Nederlands Rijpaarden en Pony Stamboek) gelding was presented to the referring veterinarian with a mass in the soft tissues caudo-dorsal to the left eye. The swelling showed rapid growth over the previous 3 weeks according to the owner. No other abnormalities, such as exophthalmos, were noted.

Ultrasonography of the swelling was performed and a fine needle aspirate was taken for cytology and culture. Cytology showed erythrocytes, macrophages and erythrophagocytosis compatible with cystic fluid and no bacteria were cultured. No definite diagnosis was made based on these examinations, but a cyst was suspected. Therefore, the pony was referred for computed tomographic (CT) imaging of the head to the Department of Equine Sciences of Utrecht University.

Clinical findings

At presentation at the Department of Equine Sciences, the pony was bright and alert and showed no signs of discomfort, exophthalmos or epiphora. No nasal discharge or abnormal odour was noted at presentation. General clinical examination and ophthalmic examination showed no abnormalities. There was a fluctuating mass in the retrobulbar area of the left eye (5 × 2 cm). The swelling could be palpated without evoking a pain reaction (**Fig 1**).

Imaging

A CT examination of the head was performed in the standing sedated horse. Sedation consisted of detomidine hydrochloride (Domosedan¹ 0.01 mg/kg bwt) administered through a catheter (Intraflon 14 G × 80 mm) placed in the right jugular vein. The CT examination was performed with 140 kVp, 302 mAs and 512 × 512 matrix and FOV adjusted to patient head size (359 mm) (Somatom Definition AS 64-slice sliding gantry)². Bone (H60f) and soft-tissue (H31f) algorithm images were reconstructed with 1 mm slice thickness. A large space-occupying mass lesion (approximately 6.3 cm height, 3.6 cm width and 7.0 cm length) was present between the left lateral aspect of the calvarium and the temporal muscle at the caudo-dorsal aspect of the ocular structures. The lesion was of mixed attenuation with a hypoattenuating centre (22 HU). Eccentric dislocation of the temporal musculature was seen as was slight deformation of the ocular musculature and retrobulbar fat with very minimal lateral exophthalmos. Moderate indentation of the lateral part of the frontal bone into the lateral part of the frontal sinus due to pressure necrosis remodelling was present. The mass lesion was continuous into the squamous part of the temporal bone causing a cyst-like structure with an opening at the rostral aspect. No evidence of sinusitis was seen on the CT examination.



Fig 1: Preoperative picture of the pony, notice the retrobulbar swelling of the left eye.

The mass lesion was considered to be compatible with an atypical bone cyst or sinonasal cyst, however, a cystic soft-tissue neoplasm could not be ruled out (**Fig 2**). Due to the invasive character of the cyst and the minimal lateral exophthalmos, the care and treatment of the patient was transferred from the referring veterinarian to the Surgical Department of the Department of Equine Sciences. In order to further define the diagnosis and to develop a treatment plan, ultrasonography, bacteriologic culture and cytology was performed. The ultrasound examination confirmed a cyst-like structure containing echoic fluid with a diameter of 5 cm. The thickness of the capsule varied between 2 and 3 mm. The cyst-like structure was continuous caudally with a cyst-like mass of the temporal bone. Under ultrasound guidance a fine needle aspirate was taken for cytology and culture. The fluid obtained was haemorrhagic and cloudy. Cytology showed erythrocytes and macrophages with erythrophagocytosis. Bacteriologic culture was negative.

Treatment

Surgery

Surgical excision of the cyst-like structure under general anaesthesia was recommended to the owner. Premedication consisted of detomidine hydrochloride (Domosedan¹ 0.01 mg/kg bwt intravenous [i.v.]) and butorphanol (Dolorex³ 0.02 mg/kg bwt i.v.). The pony also received meloxicam (Metacam⁴ 0.6 mg/kg bwt i.v.), benzylpenicillin (Benzylpenicilline Natrium⁵ 20.000 IE/kg bwt i.v.) and gentamycin (Gentamycine 5%⁵ 6.6 mg/kg bwt i.v.). The pony was induced with diazepam (Diazepam CF⁶ 0.05 mg/kg bwt i.v.) and ketamine (Narketan¹ 2 mg/kg bwt i.v.) and was kept under general anaesthesia with isoflurane 1–2% via a 22 mm

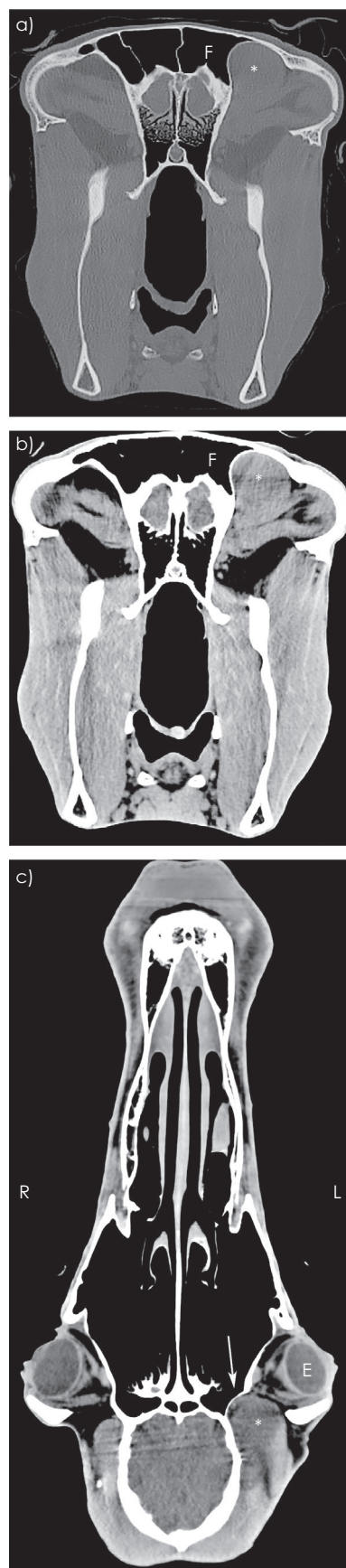


Fig 2: CT images of the head at the level of the eyes. a) bone algorithm and b) soft tissue algorithm: notice the large space-occupying mass caudo-dorsal to the ocular structures and its deforming character into the frontal sinus. c) Dorsal multi-planar reformatted image, soft tissue algorithm. Notice the dislocation of the ocular musculature and mild exophthalmos. L = left, R = right, * = dermoid cyst, E = eye, F = frontal sinus.

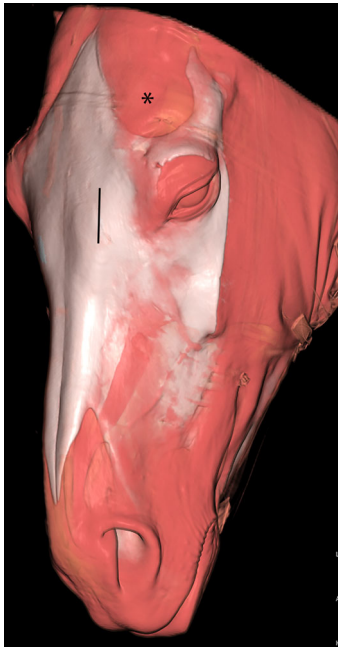


Fig 3: 3D reconstruction of the overview of the skull. Notice the soft tissue swelling caudo-dorsal to the left eye. The first surgical incision is indicated with a *. The bone flap that was eventually created to completely remove the dermoid cyst is indicated by the black line.

endotracheal tube, and detomidine hydrochloride (Domosedan¹ 0.01 mg/kg bwt/h i.v.) as a constant rate infusion. The pony was positioned in right lateral recumbency. The soft tissue swelling was present in the retrobulbar area and showed indentation into the frontal bone. However, since the frontal bone was still intact the first surgical approach was an incision in the retrobulbar area (**Fig 3**). Caudo-dorsal to the left eye a semicircular incision of 10 cm in diameter was made with a scalpel blade number 22. Retrobulbar fat was exposed and the temporal muscle was visible. Underneath the temporal muscle the cyst was palpable as a round shaped structure 5–8 cm in diameter. It was surgically impossible to remove the cyst as a whole through the aforementioned incision. A bone flap was made in the frontal sinus by performing an osteotomy with an oscillating bone saw (**Fig 3**). An outpouching of the mass in the frontal bone was palpated and an opening was made from the sinus to the retrobulbar area using a chisel and a hammer. Using this approach, we were able to completely remove the cyst from the bone through blunt dissection and the cyst was removed in toto. A thin seton drain was placed from the sinus to the retrobulbar space and was anchored with sutures to the skin 5 cm above the eye. The sinus was flushed thoroughly and the bone flap was closed in two layers. The periosteum was closed with a continuous suture pattern using Poliglecaprone 25 (Monocryl[®]). The subcutis and skin were closed with a two-layer closure, vertical mattress suture pattern using a Polyamid 6 (Ethilon[®]). A balloon catheter (size 18, 6 mm in diameter, 5–15 mL) was placed in the frontal sinus. The incision in the retrobulbar area was closed with vertical mattress sutures using Polyamid 6.

Post-operative treatment

Following surgery, the frontal sinus was flushed once a day with 0.01% povidone iodine solution for 6 consecutive days because a communication was made to the frontal sinus and a possible iatrogenic sinusitis could have been induced. After this the balloon catheter was removed from the sinus. The seton drain was left in place for 22 days, to create a permanent opening between the previously formed cavity of the cyst and the sinus. This was to prevent possible buildup of fluid in the future and therefore prevent recurrence. Facial pain scores according to the Equine Utrecht University scale for facial assessment of pain (Van Loon and van Dierendonck 2015) were recorded during 4 days post-operatively. Initially the pony only received meloxicam for 6 days (Metacam⁴ 0.6 mg/kg bwt per os once daily), however, since the pony showed signs of discomfort based on the facial pain scores (FPS 3, nostril flaring, teeth grinding and moaning), oral tramadol (Tramagetic 300⁷ 5 mg/kg bwt per os) was added twice a day for 3 days. The FPS dropped to 1. Procaine benzylpenicillin (Procapen⁹ 20 mg/kg bwt i.m.) was continued for 6 days post-operatively once a day. Mild swelling of the surgical site was seen 2 days after surgery. Mild signs of headshaking were seen during hospitalisation when the pony was turned out into a small pasture in bright light. The pony was discharged 8 days after surgery without any signs of discomfort.

Histopathology

On histopathology, the excised mass was characterised by a fibrous capsule-like oval structure, partially lined with well-differentiated keratinised stratified squamous epithelium occasionally forming papilliferous projections into the lumen of the cyst (**Fig 4**). In the pericapsular stroma, variably sized aggregates of moderate to large numbers keratinocytes, several hair fragments and fewer clear acicular clefts (cholesterol clefts) were noted, surrounded by numerous macrophages and limited numbers of eosinophils, lymphocytes and plasma cells (**Fig 5**). The histopathology is consistent with a dermoid cyst with granulomatous inflammation.

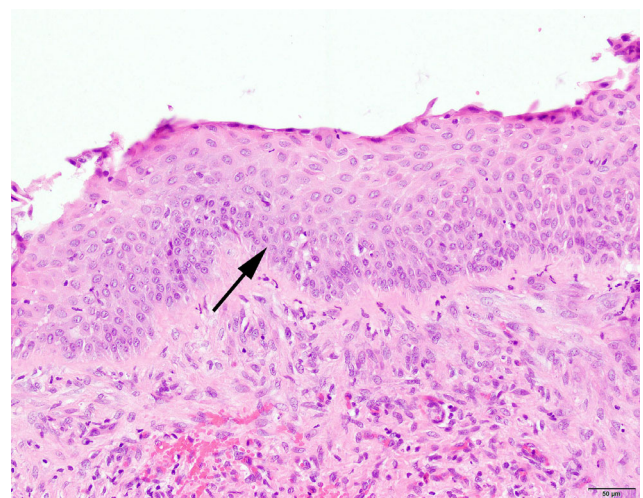


Fig 4: Well-differentiated keratinised stratified squamous epithelium (arrow). HE staining ($\times 20$).

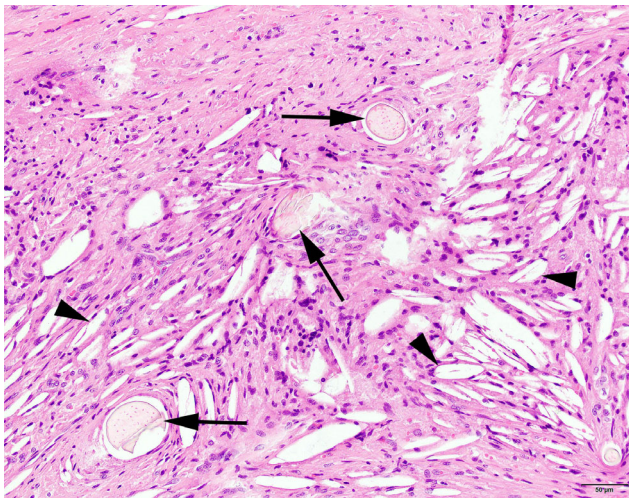


Fig 5: Granulomatous inflammation, arrows hair fragments, arrow heads cholesterol clefts. HE staining ($\times 20$).

Follow-up and outcome

Two weeks after discharge, the seton drain was removed as well as the skin sutures. No retrolbulbar swelling was present at this time. However, the owner mentioned that the pony still showed signs of headshaking in bright light in the pasture, but the owner did not desire any further diagnostics. The headshaking persisted until 2 months after surgery. When the owner was contacted for a brief follow-up at 6 months she mentioned she did not notice the headshaking at that point in time. One year post-operatively the pony is back in training and performs at his intended level of use. However, the pony shows a slight recurrence of the headshaking only in bright light at this point in time according to the owner.

Discussion

This case report describes the CT images and histopathologic diagnosis of a dermoid cyst in the retrolbulbar area of an 18-year-old pony with indentation of the frontal bone into the frontal sinus. The pony was referred with a nonspecific fluctuating mass in the retrolbulbar space of the left eye. Differential diagnoses included retrolbulbar or orbital neoplasia, orbital cellulitis and abscess, sinus cyst, hydatid cyst, myositis of the extraocular or masticatory muscles, retrolbulbar haematoma, zygomatic salivary gland mucocele or congenital malformations (Knottenbelt *et al.* 2015).

To the authors' knowledge, CT has not been described in other cases with a dermoid cyst in the retrolbulbar area of horses. In this case, a standing CT scan of the equine head was made because ultrasonography, cytology and culture performed by the referring veterinarian were inconclusive and the pony was solely referred for CT imaging. To work-up a case like this, our approach would normally be to perform an ultrasound examination of the area and perform cytology before continuing with other diagnostics. CT gave detailed information about the location and the different structures involved in order to make a complete surgical plan. The CT did confirm a retrolbulbar mass compatible with a cyst or abscess, however neoplasia could not be ruled out. Histopathologic examination after surgical removal gave a final diagnosis of a dermoid cyst.

Dermoid cysts are considered to be developmental anomalies, benign in nature and are often congenital and possibly hereditary (Scott and Miller 2011). They are considered to be the result of an entrapment of ectodermal and/or dermal elements during the closure of the neural groove (Engler *et al.* 2016; Kosuge *et al.* 2016). However, they may also have an acquired origin and arise following a traumatic disruption of epithelial structures (Muñoz *et al.* 2007; Knottenbelt *et al.* 2015). The pony described in this case was 18 years of age and swelling was only apparent for several weeks. The sudden onset and the age of the pony might suggest an acquired origin instead of a congenital origin, however, no trauma was mentioned in the clinical history.

As a differential histopathologic diagnosis an epidermoid cyst should be taken into consideration. An epidermoid cyst is also benign in nature and histopathological examination shows squamous epithelial lining with a fibrous wall and no adnexal structures. Dermoid cysts, however, do include adnexal structures and contain a central cavity filled with keratin, skin appendages and hair shafts (Sahoo *et al.* 2015; Knottenbelt *et al.* 2015).

The growth pattern of the cyst, showing indentation into the lateral part of the frontal sinus is not readily explained. Craniofacial dermoids in children have been reported to rarely extend intracranially (Reissis *et al.* 2014). A study performed by Reissis *et al.* (2014) showed no histological differences between cysts that extend intracranially and cysts that do not. Failure of cranial structures to fuse completely is presumed to be a cause of the intracranial extension and not the functional characteristics of the cyst itself (Reissis *et al.* 2014). This case report describes an older horse, which makes a congenital origin less likely. Although no trauma was mentioned in the clinical history, disruption of epithelial structures and the following pressure necrosis on the frontal sinus may be the most plausible explanation for the origin and growth pattern of the dermoid cyst.

Surgical excision is considered to be the treatment option for orbitofacial dermoid cysts in children (Yan and Low 2016). In humans, the space occupying effects and the possible intracranial extension make surgery the definitive treatment choice (Hillyer *et al.* 2003; Yan and Low 2016). However, dermoid cysts in horses seem to appear without clinical significance and tend to be asymptomatic. In horses, one could debate the need for surgical excision depending on the location of the cyst. In this case the dermoid cyst was located in the retrolbulbar space. No exophthalmos was seen clinically, however, the CT scan revealed a very mild lateral displacement of the left eye. Therefore, CT imaging in this case was of great value and provided the information needed to consider and develop a surgical plan. Another consideration for surgery was the growth rate of the cyst. The swelling had appeared to grow rapidly over the previous 3 weeks according to the owner. Although there was no evidence for this on histopathology.

In case of surgical excision, special attention should be paid to complete removal of the cystic wall. In another case report of a dermoid cyst in the retrolbulbar space described by Muñoz *et al.* (2007) retrolbulbar swelling reappeared 2 weeks after surgery. Incomplete removal of the cystic wall was a possible explanation for this reoccurrence (Muñoz *et al.* 2007). This was not the case in our patient as the complete cyst was removed during surgery. Furthermore, a seton drain was placed for a period of 22 days in order to drain possible remaining cystic fluid. Moreover, the seton drain also ensures

that the surgical opening made between the cystic cavity and the frontal sinus would not close prematurely and therefore hoping to prevent recurrence of cystic fluid buildup. No reoccurrence has appeared 1 year after treatment.

Post-operatively the pony did show a period of headshaking, despite pain medication. A possible explanation could be a trigeminal neuropathy due to the surgery. One year after surgery the pony only occasionally shows mild signs of headshaking in bright light according to the owner. A possible explanation for the absence of signs of headshaking in the period between 2 months after surgery until 1 year after surgery could be due to less bright light during winter and shorter turnout of the horses in the field. Surgery was performed in the middle of summer and the recovery after surgery mainly took place during the fall and winter. In the Northern hemisphere, where this case occurred, sun hours are limited in fall and winter. Therefore, it could be possible that the owner did not see clear signs of headshaking, although they might have been present in a mild form. Headshaking in horses can have multiple causes varying from sinus disease, dental disease to trigeminal-mediated headshaking (Pickels *et al.* 2014; Ross *et al.* 2018).

In our case, trigeminal-mediated headshaking was suspected, as the pony showed mild headshaking only in bright light, making photic headshaking most likely (Madigan *et al.* 1995; Pickels *et al.* 2014). No sinusitis was apparent pre- or post-operatively and no sinus or dental disease was noted on CT examination at time of referral that could provide an alternate explanation for the headshaking. As the pony was not seen by us at follow-up nor had a diagnostic workup performed for headshaking this was not confirmed. Headshaking was recorded in 5 horses after paranasal sinus surgery, suspecting trigeminal neuritis (Gilsenan *et al.* 2014). In 2 out of these 5 horses infraorbital canal damage occurred during surgery and in the other cases trigeminal nerve injury was suspected due to concussive trauma to the infraorbital canal during surgery. In our case, no damage to the infraorbital canal happened during surgery and the cyst was not situated close to the infraorbital canal either. Although no diagnostic imaging was performed after surgery to assess the integrity of the infraorbital canal, we do not expect the infraorbital canal to be involved in this case. A more plausible explanation might be damage to one of the sensory nerves in the retrobulbar area during surgery, causing neuropathic pain and neuropathic pain (Gilsenan *et al.* 2014). Auricular nerve branches are present in the retrobulbar as part of the facial nerve and the supraorbital nerve, a branch of the frontal nerve, which both serve a sensory task and therefore causing neuropathic pain.

Authors' declaration of interests

No conflicts of interest have been declared.

Ethical animal research

Ethical review not applicable for this retrospective case report.

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Authorship

E.M.S. Visser managed the clinical case and prepared the manuscript. S. Veraa evaluated and described the computed tomography findings. N. Caliskan performed the post-mortem and contributed to the description of necropsy findings. H. Hermans managed the clinical case and edited the manuscript. All authors approved the final manuscript.

Manufacturers' addresses

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- ⁵Dechra Pharmaceuticals PLC, Northwich, Cheshire, UK.
- ⁶Centrafarm Services BV, Etten-Leur, The Netherlands.
- ⁸Ethicon, Dülmen, Germany.
- ⁷Bard Pharmaceuticals Limited Cambridge Science Park, Cambridge, UK.
- ⁹aniMedica GmbH, Senden-Bösensell, Germany.

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