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CASE REPORT

Surgical treatment of a five-structure dentigerous cyst in a Warmblood filly

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Summary

This article describes a unilateral extracranial dentigerous cyst containing five dental structures in a 3-year-old Warmblood filly that caused drainage from below the left pinna and behavioural changes due to pain. Initial diagnosis was made using radiographs and further diagnostic imaging in the form of computed tomography was used for effective surgical planning. This highlighted the proximity of the cyst to the calvarium. All the structures were surgically removed using an osteotome and mallet. During surgery, a defect was made in the calvarium leading to a small haemorrhage, which was successfully controlled. The external auditory canal was accidentally entered during the procedure; closure was achieved and no postoperative infection was experienced. The structures were histologically confirmed to be dental in origin. On telephone and photographic follow-up, the horse showed excellent cosmetic results, with no neurological or motor deficits and complete resolution of clinical signs.

KEYWORDS
horse, computed tomography, dental, surgery

INTRODUCTION

Dentigerous cysts, also named temporal teratomas or heterotopic polyodontia, are a relatively common congenital abnormality described in young horses (Mason, 1974). They form due to ectomesenchymal cells failing to migrate to the correct position in the maxillary or mandibular arcade from the midbrain and hindbrain (Cobourne & Sharpe, 2003). They become fixed and are normally pushed up towards the temporal region, due to failure of the first branchial cleft to close (Jubb et al., 2007). The majority of dentigerous cysts are located in the temporal region; most commonly associated with the external pinna. They have also been described within the ventral nasal meatus (De Mira et al., 2007) and ventral conchal sinus (McClure et al., 1993). Most cysts are unilateral, although bilateral presentation has been reported (Smith et al., 2012).

Dentigerous cysts normally present as a soft swelling below the base of the pinna with an associated draining tract (Mason, 1974). Cysts in horses can be described as dermoid (not containing a tooth), or dentigerous (containing a tooth) (Misdorp, 2003). This differs from dentigerous cysts in humans, which are unerupted or impacted teeth in the mandible or the maxilla (Zhang et al., 2010).

The fluid produced contains desquamated stratified squamous epithelial cells (Brown et al., 2007). The continuous proliferation of these cells causes the cysts to grow over time. The swelling can be non-painful and treatment is elected for cosmetic reasons in many cases. Secondary infection of the fluid within the cyst can lead to pain and irritation (Brown et al., 2007; Easley et al., 2010; Mueller, 1991). Medical management using antibiotics or drainage of the cyst is unrewarding and the treatment of choice is surgical excision (Rashmir-Raven et al., 1990). Diagnostic imaging is recommended prior to surgery to determine the exact location and extent of the cyst and its proximity to important anatomical structures such as the temporomandibular joint and the calvarium (Easley et al., 2010; Hunt et al., 1991).

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CASE PRESENTATION

A 3-year-old Warmblood filly presented with swelling and mucopurulent discharge ventral to the left ear. The owner reported that the filly had become sensitive and head shy on the left-hand side over a period of a few months and was refusing to be bridled. The swelling was minimal, and the discharge had ceased when the filly was first seen by the attending veterinarian. Under standing sedation, the sinus tract was surgically debrided due to suspicion of localised infection and was sutured closed. A single dose of procaine penicillin and a single dose of tetanus antitoxin were given preoperatively. Five months later the swelling and mucous discharge recurred. At this time radiographs were obtained and a diagnosis of a large dentigerous cyst (Figure 1) was made by the attending veterinarian before referring the case to the Equine Veterinary Clinic of Massey University for surgical excision.

CLINICAL FINDINGS

The filly was presented with significant swelling ventral and rostral to the left ear. A sinus tract with minimal dried exudate at its cloaca was observed (Figure 2). The filly was extremely reactive to palpation of the left temporal region and was evasive when handled from the left-hand side. A neurological examination was performed, and the filly did not display any central or peripheral neurological deficits.

DIAGNOSTIC IMAGING

Due to the extent of the cyst and suspected proximity to the brain, it was elected to perform computed tomography (CT) of the head for accurate surgical planning. This was done with the horse under general anaesthesia in dorsal recumbency. The filly was premedicated with acepromazine (0.03 mg/kg bwt IV) and xylazine (0.4 mg/kg bwt IV). Induction was performed with ketamine (2.2 mg/kg bwt IV) and diazepam (0.1 mg/kg bwt IV) and anaesthesia was maintained using propofol continuous rate infusion (CRI). Oxygen was provided via a demand valve and orotracheal tube.

Computed tomography images were obtained with a slice thickness of 2.0 mm with 1.0 mm overlap and the tube output was 120kVp and 242 mA. The study was acquired in a transverse plane using a bone algorithm (YD filter) and reconstructions were made using a soft tissue algorithm (B Filter). Plain views were obtained first and then a fistulogram was attempted using a 14G catheter and iohexol (300 mg/ml). High resistance was encountered upon injection and only a small amount of contrast could be injected, none of which appeared to reach the cyst.

The CT showed the presence of five dental structures within the left temporal and occipital bones, some of which resembled complete molar teeth, while others were small and amorphous. The bone surrounding these structures had an expansile appearance. The left occipital bone was distorted and the structure severely impinged upon the left occipital lobe (Figure 3). The calvarium appeared intact in between the cyst and the brain itself. On the soft tissue reconstruction there did not appear to be any abnormality to the brain tissue itself. There was also distortion of the petrosal portion of the left temporal bone by the dental structures. The structures were included in the squamous portion of the temporal bone. The left external ear canal was displaced ventrally and markedly narrowed, without fluid accumulation in the tympanic cavity (Figure 4). The mastoid process, tympanic cavity and temporomandibular articulation (Figure 5) had mild distortion.

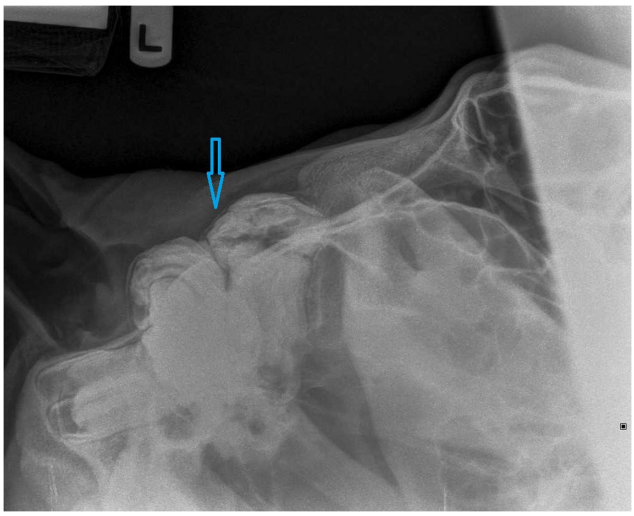

FIGURE 1 Rostral 70° lateral 45° ventral-caudodorsal oblique radiographic projection of the left temporal region highlighting the presence of at least three clearly identifiable dental structures (blue arrow) at the base of the left ear. Rostral is to the right of the image and dorsal is to the top.

TREATMENT

Based on the extent of the cyst and the proximity of the dental structures to the calvarium, it was elected to perform surgical removal under general anaesthesia 2 days after the CT. Preoperative phenylbutazone (2.2 mg/kg bwt IV), gentamicin (6.6 mg/kg bwt IV) and procaine penicillin (22 mg/kg bwt IM) were administered. The filly was premedicated with butorphanol (0.01 mg/kg bwt IV) and detomidine (0.01 mg/kg bwt IV). Anaesthesia was induced with ketamine and diazepam and maintained with inhalation of isoflurane in oxygen. Lidocaine was administered intravenously as a CRI of 0.05 mg/kg/min for pain control and to reduce the minimal alveolar concentration requirement for isoflurane. Morphine (0.1 mg/kg bwt IV) was given at the start of surgery and again in recovery. The horse was positioned in right lateral recumbency, the area around the base of the left pinna was clipped and aseptically prepared with the pinna wrapped in a sterile adhesive material to prevent contamination.

An approximately 10 cm long slightly curved incision was made ventral and rostral to the medial ridge of the left pinna and through the cloaca of the sinus tract. The soft tissues were bluntly dissected until calcified tissue was encountered. A proper cyst lining could not be identified by following the sinus tract nor by exploring the base of the pinna where an opening is generally present with dentigerous

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FIGURE 2 An obvious swelling is visible at the base of the left pinna. Dried exudate is present in the middle of the swelling, occluding the cloaca.

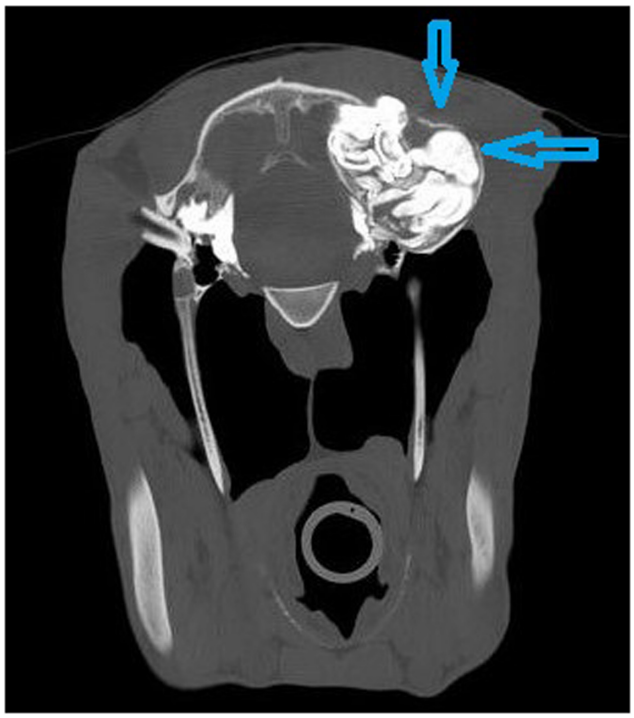

FIGURE 3 Computed tomography transverse image in the temporal region (window width of 2500 and window level of 500). Left is to the right of the image, dorsal is to the top. Notice the extent of the dentigerous cyst (blue arrows) in the left temporal region of the skull compressing the left occipital lobe of the brain.

FIGURE 4 Computed tomography transverse image at the level of the external ear canal (window width of 2500 and window level of 500). Left is to the right of the image, dorsal is to the top. Note the ventral displacement and marked narrowing of the left external ear canal (blue arrow), without fluid accumulation in the tympanic cavity.

cysts. The dental structures were firmly attached and could not be loosened from the surrounding bone by prying with osteotomes. It was elected to break the mass into pieces with an osteotome and mallet. The skin incision was extended along the medial aspect of the pinna for an additional 6–7 cm in order to create a skin flap (Figure 6). The structures deep to the first teeth encountered were adherent to the calvarium and were freed carefully by systematically loosening their attachment to the bone.

The external auditory canal was accidentally entered during the surgery and was sutured closed using 2–0 poliglecaprone 25. All of the dental structures were extracranial and firmly attached to the calvarium. During removal of the deep dental structures a small defect (5 × 5 mm) was inadvertently made in the calvarium. This resulted in mild venous haemorrhage with an estimated blood loss of 500 ml. The haemorrhage could not be initially stopped by applying pressure with plain gauze, but a cellulose-based haemostatic dressing (Woundclot™ Core Scientific Creations) provided effective haemostasis. Intraoperative radiographs were taken to guide the surgical procedure and to confirm complete removal of the mass. All dental structures removed weighed 288 g (Figure 7).

A Penrose drain was placed in the cavity and was exited through a separate incision created on the lateroventral portion of the temporal region, and the surgical incision was closer in three layers. The anaesthetic time from induction to placement of the filly in the recovery room was three and a half hours.

POST-OPERATIVE CARE

After recovery from anaesthesia, the filly developed right facial nerve paralysis and swelling of the right masseter muscle and showed

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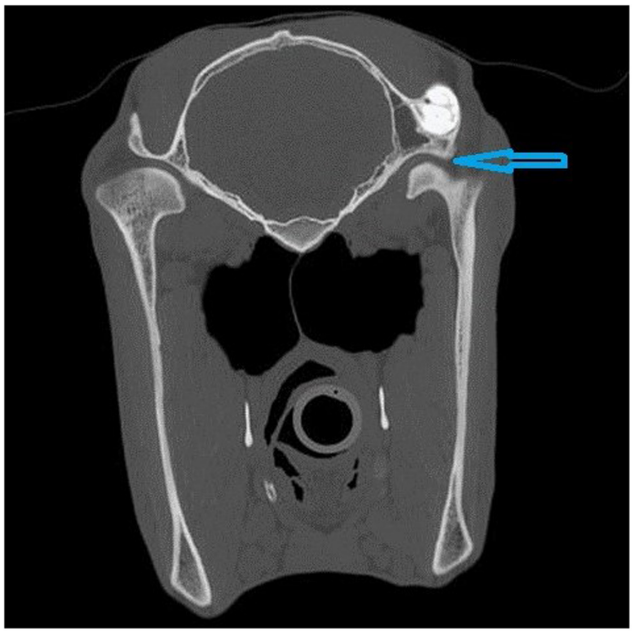

FIGURE 5 Computed tomography transverse image at the level of the temporomandibular joint (window width of 2500 and window level of 500). Left is to the right of the image, dorsal is to the top. Note the presence of a dental structure in close proximity to the temporomandibular joint (blue arrow).

FIGURE 6 Location and extension of the surgical incision (photograph taken 4 days after surgery)

right forelimb lameness with a gait consistent with suprascapular neuropraxis. These signs were on the dependent side and were suspected to be due to prolonged lateral recumbency. On neurological

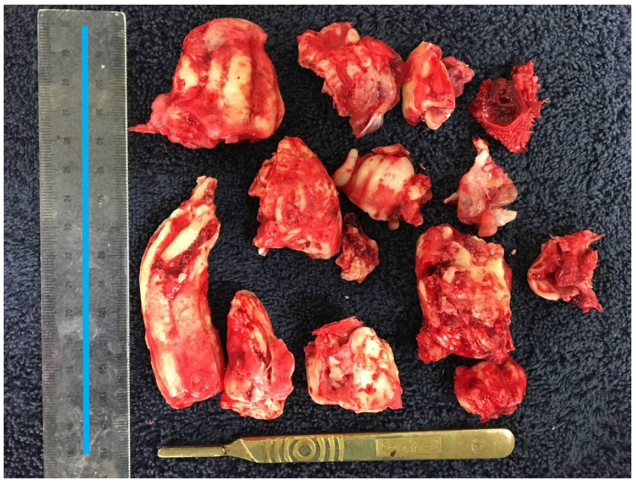

FIGURE 7 Dental structures after surgical removal. The molariform structure at the bottom left was the most caudally and dorsally located into the occipital bone, medial to the left pinna. This measured 8 cm in length. The blue line on the ruler measures 15 cm and provides a reference scale.

examination no peripheral neurological signs were observed in the left temporal and auricular regions suggesting there was no nerve deficit due to the surgery itself. The left ear also showed good mobility and was responsive to auditory stimuli. Vision and central nervous functions were considered normal.

Post-operatively the filly was kept on phenylbutazone for 7 days and gentamicin and procaine penicillin for 5 days due to the potential risk of meningitis associated with infection from the external auditory canal or the small defect made in the calvarium. The day after surgery, creatine kinase (CK) was found to be elevated at 7867 IU/L (110–250 IU/L). Creatinine was found to be normal at 120 μmol/L (85–165 μmol/L). The filly was kept on twice maintenance IV Hartman's fluids overnight to reduce the risk of renal damage from the elevated CK. Intravenous lidocaine CRI (0.05 mg/kg/min IV) was administered for 18 h post-operatively. This was given to minimise post-operative pain and risk of colic. The facial paralysis and gait significantly improved in the following 7 days. The area around the Penrose drain was cleaned every 6 h; minimal serous discharge was produced so the drain was removed 4 days post-operatively. During hospitalisation, the filly was monitored with physical examinations every 6 h with particular attention to rectal temperature and mentation due to potential complications of meningitis or local infection. After 7 days, the filly was discharged from the hospital and the staples were removed 2 weeks post surgery.

HISTOPATHOLOGY

The tooth-like structures contained areas of stellate reticulum lined with enamel epithelium and enamel formation overlying dentine with prominent dentinal tubules. Dental pulp was also present in some of the structures. The teeth varied from amorphous to normal dental architecture. Sections through the periphery of larger teeth

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showed more normal architecture. This confirmed the structures to be dental in origin.

FOLLOW-UP

A telephone follow-up with the owner was carried out 4 months post-operatively. The filly showed no neurological or visual deficit on the left side, and the head shyness associated with this side was almost resolved. The facial and suprascapular nerve deficits on the right side were fully resolved by 3 weeks after surgery. The surgical site had healed well without swelling or discharge, with only a small fibrous lump palpable. The filly was now working well under saddle. Twenty-six months post-operatively the horse had been competing well in showjumping and won two prizes. The owner reported no further clinical signs associated with the cyst and the head shyness was fully resolved.

DISCUSSION

Dentigerous cysts are most commonly described as containing only one dental structure. Nevertheless, multiple structures have been described (Easley et al., 2010; Gaughan, 2010; Hunt et al., 1991; Mason, 1974), only one other study has previously documented five structures (Cronau et al., 2009). The extent of the cyst described in this report caused severe distortion of the occipital bone with consequent compression of the occipital lobe of the brain. There is no previous description of a cyst causing this degree of anatomical deformation being removed successfully. CT or magnetic resonance imaging have previously been described as the gold standard for imaging prior to surgical removal as they allow the best 3D visualisation of the area (Smith et al., 2012). In this case, CT imaging was extremely valuable for surgical planning as well as for an informed discussion with the client about the potentially fatal complications of surgery given the risk of breaching the calvarium. Structures located intracranially and in direct contact with the brain would carry a much poorer prognosis for safe surgical excision (Cronau et al., 2009).

Surgical removal is the most effective form of treatment and requires careful dissection of the entire capsule (Rashmir-Raven et al., 1990). Potential complications of surgery include profuse haemorrhage, meningitis, concussion, fracture, damage to the external auditory meatus and damage to the nerves and muscles of the ear, causing paralysis of the pinna or other areas of the face (Carr, 2006). Given the proximity to the brain in this filly, there was also high risk of entering the calvarium as described by others (Cronau et al., 2009; Hunt et al., 1991) and death could have occurred (Cronau et al., 2009). In the present report, a small full thickness defect was inadvertently made in the calvarium, which is believed to be the cause of the intraoperative haemorrhage; this did not appear to have a negative effect on the recovery of the filly. Other structures that could be inadvertently entered include the temporomandibular joint, potentially leading to sepsis and the external auditory canal (Smith et al., 2012). The latter was entered during surgery in this filly but was sutured closed immediately and broad-spectrum antibiotics were continued for 5 days postoperatively due to presumed contamination of the surgical site from the non-sterile external auditory canal. A major concern with any contamination in proximity to the brain is meningitis. The filly was monitored closely after surgery and displayed no clinical signs of meningitis. The filly also experienced right facial and suprascapular neuropraxia after surgery. These complications are generally associated with lateral recumbency and are most likely related to uneven pressure due to positioning aids in this case. As the CK was elevated and there was swelling of the right masseter muscle, a degree of masseter myositis was also suspected, which again was deemed to be related to suboptimal positioning in lateral recumbency for this filly. These complications resolved within 3 weeks post surgery.

In humans the occipital lobe is responsible for visuospatial processing, discrimination of movement and colour (Wesmoreland et al., 1994). If damage occurs to these areas, vision deficits and visual hallucinations can occur. These changes cannot be easily documented in horses, and at best could be shown by 'spookiness' or jumping at approaching objects or people. These signs were not reported in this filly pre- or post-operatively. A possible explanation for the lack of neurological signs in our case could be due to the fact that the cyst severely impinged only on the caudal portion of the left occipital lobe, thereby causing limited damage, if any. A slight impairment though may have been difficult to notice given that horses are not required to perform fine tasks like humans. Another possible reason for the lack of neurological signs could be related to the congenital nature of the condition. In fact, the cyst would have likely grown at a similar rate as the brain in the development stages early in life. It is plausible that neuroplasticity in a young animal can allow for significant adaptation, with other areas of the brain compensating for the ones being compressed or damaged.

Cysts are not historically described to be painful and removal is often elected for based on cosmetic reasons (Easley et al., 2010; Mason, 1974; Mueller, 1991). However, they are prone to infection and, when this occurs, they usually result in pain and irritation in the area (Brown et al., 2007). In this case, despite the potential for life-threatening complications, the decision to remove the cyst was made since repeated infections were causing discomfort and evasive behaviour on the left-hand side of the head. This was resulting in the deterioration of the behaviour and quality of life of the filly. In conclusion, dentigerous cysts containing multiple dental structures in close proximity to the calvarium can be removed successfully using standard surgical techniques, with the aid of CT imaging and careful surgical planning. Given the potential complications of this procedure including brain damage and death, assessment of the impact of the cyst on the horse's welfare and informed owner consent are critical before deciding to proceed.

AUTHOR CONTRIBUTIONS

All three authors were involved in the case, concept of the paper and on-going management of the patient. First opinion and follow-up

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was carried out by E. Simpson and surgical referral treatment by A. Howell and L. Panizzi. All authors contributed to the preparation of the manuscript and gave their final approval of the manuscript.

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CONFLICT OF INTEREST

No conflicts of interest have been declared.

ETHICS STATEMENT

No specific approval from local or national bodies was needed for this study. We had full permission from the owner of this horse to use the information and figures included in the paper. The owner has been kept informed of this publication and the information used within it and client confidentiality and a high standard of veterinary care has been maintained at all times.

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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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