

# Case Report

## A novel location and *en bloc* excision of a thyroglossal duct cyst in a filly

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### Introduction

Several types of developmental cysts have been reported in the cervical region of mammals. Most originate from the branchial arches and have similar manifestation across different species (Clark *et al.* 1989; Hance *et al.* 1992; Sadler 2000). The embryonic thyroid migrates as the thyroglossal duct from the base of the tongue to its final location in the cervical region (Noden and De Lahunta 1985; Latshaw 1987). A cyst at the origin of the duct is a relatively common developmental disorder in man and is referred to as thyroglossal duct cyst while subepiglottic cysts are believed to be the equine parallel (Robertson 1991; Sadler 2000). In the horse the principle diagnosis modalities for cervical and pharyngeal cysts include upper airway endoscopy and lateral laryngeal radiographs (Embertson 1998; Davenport-Goodall 2003). Several methods have been reported for treating pharyngeal and cervical cysts. All methods attempt to eliminate all secreting epithelium while minimising damage to surrounding tissue (Tulleners 1991; Slovis *et al.* 2001).

**In the case reported here, the cyst represented the most distal end of the thyroglossal duct and was therefore unique both in its location and histological features. *En bloc* surgical excision of the cyst proved successful in this case.**

### Case details

#### Case history

A 2-day-old Thoroughbred filly was admitted to the University of Missouri Veterinary Medicine Teaching Hospital for evaluation of a cranial ventral cervical swelling. The filly

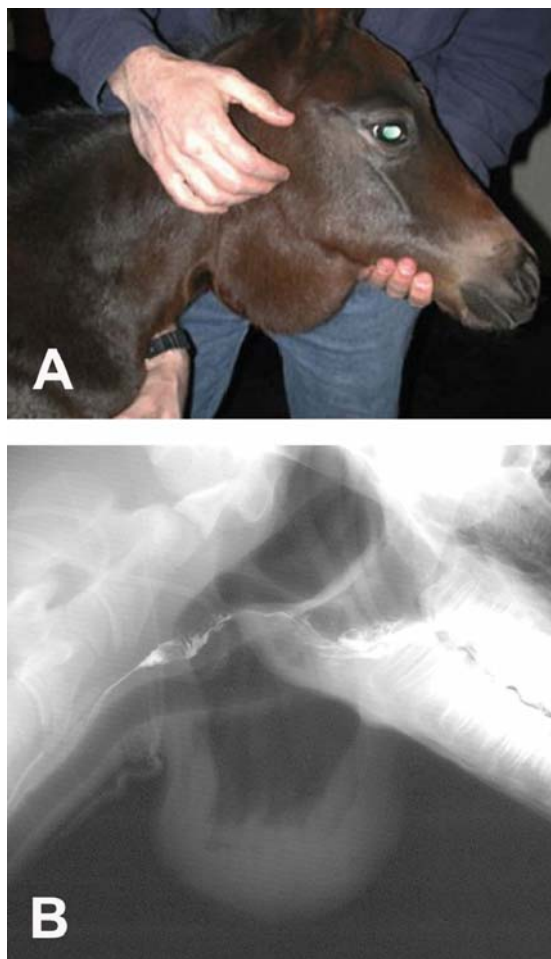
appeared normal, alert, healthy and nursed immediately following parturition. A cervical swelling was noted soon after parturition and this was drained by the referring veterinarian, who noted clear serous fluid. The next day the swelling returned and enlarged. Foul-smelling green-coloured fluid was drained from the swelling, and the filly was referred for further evaluation.

#### Clinical findings

Upon presentation the filly appeared bright and alert. The only abnormality noted upon physical examination was a 12 cm diameter swelling at the cranial midline in the ventral cervical region (**Fig 1**). Haematological abnormalities included marked leucopenia ( $2.8 \times 10^9/l$ ; reference range,  $5.4\text{--}14.3 \times 10^9/l$ ), with neutropenia ( $0.91 \times 10^9/l$ ; reference range,  $2.26\text{--}8.58 \times 10^9/l$ ). The serum IgG concentration was within normal limits (above 8.0 g/l). Blood culture was negative. The filly was placed on i.v. broad-spectrum systemic antibiotics (amikacin 21 mg/kg bwt q. 24 h and potassium penicillin 22,000 iu/kg bwt q. 6 h).

The following day the swelling was grossly enlarged and fluid-filled. Ultrasonographic evaluation of the mass revealed a nonuniform, hypoechoic fluid, consistent with an abscess or a haematoma. Aseptically aspirated fluid was discoloured and cloudy and a sample was submitted for cytology, bacterial isolation and antimicrobial susceptibility testing. Cytology of the fluid was consistent with intense suppurative inflammation. *Streptococcus equi zooepidemicus* and *Escherichia coli* were isolated. Both bacteria were susceptible to a variety of antimicrobials including penicillin, amikacin and trimethoprim-sulphadiazine. The swelling was lanced, drained and flushed with sterile isotonic fluid solution, and a formalin-fixed wedge biopsy of the wall was submitted for histopathology. Videoendoscopy of the upper airway and nasopharyngeal region was performed and ruled out communication of the cavity with the oesophagus and the trachea. Plain and contrast lateral cervical radiographs

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**Fig 1:** A) A 2-day-old Thoroughbred filly at presentation with a distinct midline cranial ventral cervical mass. B) Upper gastrointestinal contrast study, of the same filly, using barium paste. The contrast material is seen throughout the oesophagus and negates oesophageal and tracheal communication with the cyst.

confirmed these findings (**Fig 1**). A tentative diagnosis of branchial cyst was made and treatment plan included initial stabilisation followed by surgical removal. Intravenous antibiotic therapy was continued and the filly was clinically stable for several days. Subsequent to marked improvement in haematology parameters (WBC  $4.9 \times 10^9/l$  and neutrophils  $2.98 \times 10^9/l$ ) surgery was planned. Prior to surgery flunixin meglumine (0.5 mg/kg bwt i.v. q. 12 h) was administered in order to provide analgesia and decrease inflammation.

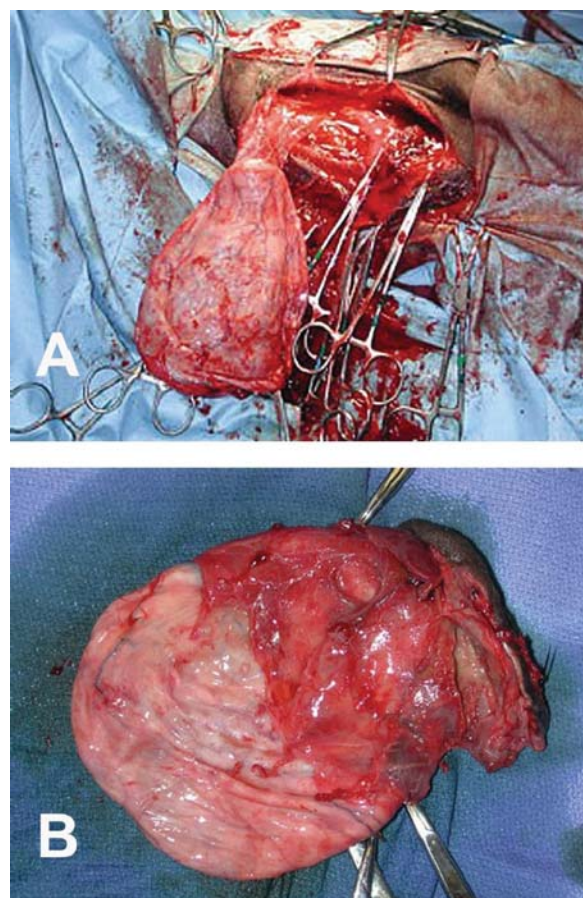
### Treatment

Surgery was performed in dorsal recumbency; the cavity was flushed with sterile balanced electrolyte solution and sutured closed prior to aseptic preparation of the surgical site. A 22 cm long fusiform incision was made over the ventral aspect of the swelling. The cyst was well defined and was removed in its entirety (**Fig 2**) along with the right lobe of the thyroid gland due to its incorporation within the cyst's wall. Removal was

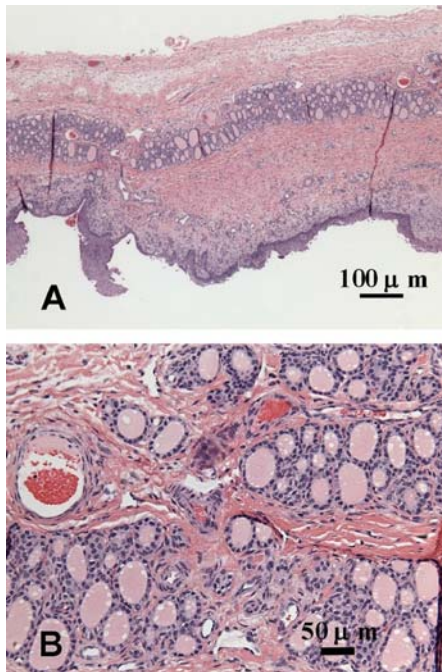
performed by combination of careful blunt and sharp dissection, and the formalin fixed mass was submitted for histopathology. Due to the previously recognised local infection, only partial closure was performed. Most of the incision was left open to allow for adequate drainage and to heal by second intention. Recovery from anaesthesia was uneventful and the filly continued to do well following surgery. No signs of dysphagia, abnormal respiratory noises or dyspnoea were noted at any time during recuperation. Following surgery, serum thyroid hormones, triiodothyronine (T3) and thyroxine (T4) levels were found to be similar to the normal reference values for a week old foal (T3 = 2.35  $\mu g/l$  [mean reference level 6.31  $\mu g/l$ ] and T4 = 63  $\mu g/l$  [mean reference level 74.5  $\mu g/l$ ]) (Irvine and Evans 1975).

### Surgical findings

Both initial and excisional biopsy results were consistent with a well defined cyst with a large central lumen. The cyst was lined by stratified squamous epithelium of varying thickness, and the wall contained follicles of varying size lined by uniform

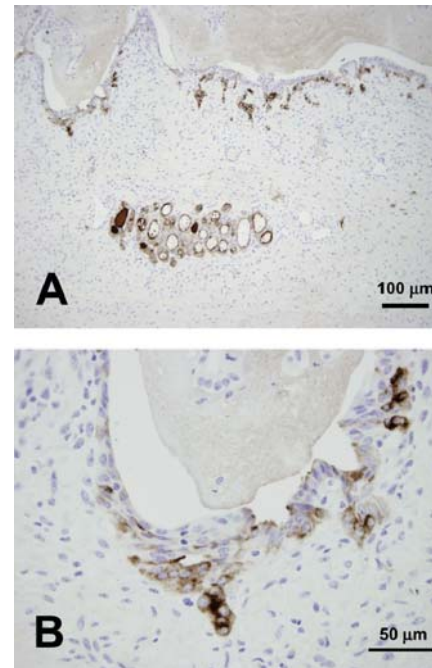


**Fig 2:** A) Intraoperative image depicting the cyst still connected by subcutaneous tissue to the thyroid region just prior to complete cyst extirpation. B) Post operative image depicting the whole cyst. The pale cystic wall on the left side shows the cyst as a distinct structure with clear separation from the subcutaneous tissue. At the far right the fusiform shaped skin removed is noted.



**Fig 3:** A) Low magnification histology slide of the cyst wall stained in H&E. The cyst is lined by stratified squamous epithelium of varying thickness, and the wall contains follicles of varying size lined by uniform layers of cuboidal cells and filled with homogeneous pink colloid. B) High magnification histology slide of the cyst wall stained in H&E. The cyst wall contains follicles of varying size lined by uniform layers of cuboidal cells and filled with homogeneous pink colloid.

layers of cuboidal cells and filled with homogeneous pink colloid (**Fig 3**). Immunohistochemical staining techniques were performed to detect expression of thyroglobulin, thyrocalcitonin and parathyroid hormone in the wall of the cyst and surrounding tissues. Positive thyroglobulin staining was present in the inner cell membrane and the cell cytoplasm of the cyst lining and adjacent follicles. The follicular content was often darkly stained (**Fig 4**). No expression of parathyroid hormone or thyrocalcitonin was observed.



**Fig 4:** A) Low magnification view of a portion of the cyst wall. Thyroglobulin immunohistochemistry, haematoxylin counter stain. Positively stained cells occur in the wall of the large cyst, and there is mild staining of luminal content at the top of the micrograph. Most of the positive cells are present in the deeper portions of the cyst wall, but some surface staining is also present. Clusters of follicles, present in the stroma away from the cyst wall, also stain positively, along with their content. B) Higher magnification of the wall of a large follicle, demonstrating staining of the epithelium of the wall.

### Outcome

The filly was sent home on oral trimethoprim-sulphadiazine (15 mg/kg bwt, *per os*, q. 12 h for 14 days) and daily wound cleansing with isotonic fluids. The filly recovered well, and the wound healed without complications (**Fig 5**). Three years following surgery the mare was in excellent health and no



**Fig 5:** A) The filly 2 weeks following surgery. The ventral cervical wound is healing well. The swelling is minimal and drainage is well maintained. B) The filly 3 years following surgery. The cervical region appears completely normal with no visible scar tissue remaining.

blemish was noted at the surgical site (Fig 5). Follow-up serum thyroid hormones testing was undertaken at 3 years of age, at which time triiodothyronine (T3) and thyroxine (T4) levels were within the normal reference range for adult horses (T3 = 700 ng/l; [reference range 250–900 g/l] and T4 = 18 µg/l [reference range 5–30 µg/l]).

## Discussion

### Establishing diagnosis

The finding of a cranial ventral cervical soft tissue swelling in a newborn foal, combined with the facts that the mass was congenital and initially yielded clear, serous looking fluid, led us to believe that the structure was a developmental cyst. Following the identification of colloid-filled thyroid follicles upon histopathology and confirmative immunohistochemical identification of thyroglobulin, a diagnosis of thyroglossal duct cyst was established. Differential diagnoses included: eustachian tube diverticulum (guttural pouch) abnormalities such as tympany or empyema, branchial cyst, ultimobranial cyst, oesophageal and tracheal duplication cysts, and salivary mucocele (Orsini *et al.* 1988; Gaughan *et al.* 1992; Hance *et al.* 1992; Sams *et al.* 1993; Peek *et al.* 1995; Slovis *et al.* 2001). Ultimobranial cyst was ruled out after the immunohistochemical stain was negative for thyrocalcitonin.

### Embryology

Pharyngeal cysts in horses are most commonly found in the subepiglottic tissue and referred to as subepiglottic cysts. There is a single report of 2 horses with soft palate cysts that were suspected to be congenital in origin (Haynes *et al.* 1990). Occasionally pharyngeal cysts appear at the dorsal pharynx. The latter are believed to be remnants of Rathke's pouch (Koch and Tate 1978; Robertson 1991). In the embryo, Rathke's pouch gives rise to the craniopharyngeal duct, which is the precursor of the anterior pituitary gland. Persistence of the pharyngeal portion of the craniopharyngeal duct is common in dogs, especially in brachiocephalic breeds and results in dorsal pharyngeal cysts. In dogs these are typically an incidental finding without clinical significance, but in the horse, related clinical signs are similar to those caused by subepiglottic cysts (Jubb and Kennedy 1970; Raker 1976; Koch and Tate 1978). The majority of the thyroid gland develops from the pharyngeal floor between 2 endodermal swellings that form the tongue. The thyroglossal duct initially indicates the line of migration of the primordial thyroid from its origin at the pharyngeal epithelium at the base of the tongue to its location in the adult at the level of the second tracheal ring. As the thyroid primordium migrates it incorporates the fifth pharyngeal pouch or the ultimobranial body, which gives rise to the parafollicular calcitonin-secreting cells (Dyce *et al.* 1987; Latshaw 1987). Ultimobranial cysts have been described in several species including dogs and ruminants (Jubb and Kennedy 1970).

### Subepiglottic cysts

In man, the thyroglossal duct is a narrow canal that is initially patent, although in most mammals the thyroglossal duct is never patent. In the course of normal development it regresses completely, and it is not found in the newborn. Thyroglossal duct cysts are well described in man. They can form anywhere along the migration path of the thyroid gland at midline but typically appear near the ducts origin by the hyoid bone (Sadler 2000). Subepiglottic cysts in horses are believed to originate from the thyroglossal duct (Colin 1973; Raker 1976; Koch and Tate 1978; Lokai and Ford 1979; Noden and De Lahunta 1985; Robertson 1991; Palmar 2003; Jones 2004). Typical histological findings of subepiglottic cysts include a variety of linings including cuboidal, stratified or pseudostratified columnar epithelium (Robertson 1991; Tulleners 1991). In a report on 3 foals, histological evidence of inflammation was found, suggestive of traumatic origin (Stick and Boles 1980). Interestingly, in a case series of 10 horses with excised subepiglottic cysts they were lined with secreting epithelium but no thyroid or thyroglossal duct tissue was identified (Tulleners 1991).

Diagnosis of subepiglottic cysts is typically made by upper airway endoscopy and lateral cervical radiographs (Tulleners 1991; Tulleners *et al.* 1999). Subepiglottic cysts typically appear in young Thoroughbred and Standardbred racehorses which may support the theory of a congenital origin (Tulleners *et al.* 1999). Occasionally, subepiglottic cysts can be found in foals. Typically, subepiglottic cysts manifest clinically in exercise intolerance and abnormal noise on inspiration like many other equine upper respiratory abnormalities (Davenport-Goodall 2003). Occasionally other clinical signs appear such as dysphagia, oesophageal obstruction, cough and nasal discharge (Koch and Tate 1978; Tulleners 1991). In 3 foals, the presenting complaint was chronic nonresponsive pneumonia. The foals were dysphagic due to the epiglottis being elevated by the subepiglottic cysts and they developed secondary aspiration pneumonia (Stick and Boles 1980). An unusual clinical presentation of subepiglottic cyst was reported by Hay *et al.* (1997). They described repeated episodes of syncope due to complete obstruction of the *rima glottidis* by a pedunculated subepiglottic cyst.

Treatment options for subepiglottic cyst include excision via laryngotomy, standing transendoscopic electrosurgical resection using a snare wire, contact or noncontact LASER (Diode or Nd-YAG) ablation, or removal and resection via an oral approach using an obstetric wire snare or LASER under general anaesthesia. The goal of surgery is complete removal of the secreting epithelial lining of the cyst while avoiding excessive removal of pharyngeal mucosa which may result in cicatrization beneath the epiglottis (Tulleners *et al.* 1999). Following surgical removal the prognosis for horses with subepiglottic cyst to return to their prior level of use is favourable (Feige *et al.* 1996; Davenport-Goodall 2003; Palmar 2003).

## Branchial cysts

Branchial cysts, which are similar in origin, structure, size and location to the current cyst, have been successfully treated both by complete surgical extirpation and by marsupialisation combined with iodine sclerotherapy (Hance *et al.* 1992; Sams *et al.* 1993; Slovis *et al.* 2001). As with any cystic structure complete removal or destruction of the epithelial lining is essential in order to prevent recurrence. Complete surgical excision is the most direct method of removal but the risk of damaging vital structure in the vicinity such as the carotid artery and the vagosympathetic trunk is high. Marsupialisation has the potential to be performed with less risk, but it entails prolonged hospitalisation and significant post operative management. Because the thyroglossal duct cyst in this filly had clearly distinguishable borders and short hospitalisation was desired by the client, we elected to remove the cyst surgically. Meticulous combined sharp and blunt dissection allowed for successful *en bloc* removal without undue difficulties.

## Conclusion

To the author's knowledge this is the first report of a thyroglossal duct cyst in the horse with supporting histological and immunohistochemical evidence showing definite thyroid tissue within the cyst wall. The location of the cyst is also unique. Most equine thyroglossal duct cysts form at the origin of the thyroglossal duct in the subepiglottic tissue. In the present case the cyst was adjacent to the thyroid at the most distal end of the thyroglossal duct. Thyroglossal duct cyst should be included as a differential diagnosis when encountering a foal with a fluid filled ventral cervical mass, and surgical excision is an effective method of treatment.

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