

CASE REPORT

Horses and other equids

Management of a mandibular sialo-cutaneous fistula in a gelding using intraluminal stenting with a polyurethane catheter

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Abstract

A 22-year-old gelding was referred to the Royal Veterinary College for evaluation of a firm swelling in the mandibular region and associated respiratory distress. Physical examination and further diagnostics, including sialography, revealed a mandibular sialo-cutaneous fistula. Taking patient and surgical factors into consideration, it was elected to place a polyurethane long-stay catheter into the duct to act as a stent and allow bypass of saliva. This technique resulted in secondary intention healing of the fistula and complete resolution of the clinical signs.

BACKGROUND

To the authors' knowledge, this is the first report of the use of a polyurethane catheter as an intraluminal stent to permit secondary intention healing of a sialo-cutaneous fistula.

CASE PRESENTATION

A 22-year-old, warmblood gelding was referred to the Royal Veterinary College Equine Referral Hospital for evaluation of marked soft tissue swellings around the left mandible of 3-week duration. The swellings had gradually increased in size and failed to respond to antimicrobial therapy prescribed by the referring veterinarian. On presentation, the gelding was tachycardic (heart rate 60 beats per minute) and tachypnoeic (respiratory rate: 32 breaths per minute), with moderate inspiratory effort and nostril flaring. Marked ptyalism was evident and the tongue protruded beyond the level of the incisors; firm swellings were palpated over the left mandible, axial to the junction of the horizontal and vertical ramus of the mandible, and between the left and right mandibles caudal to the mandibular symphysis.

INVESTIGATIONS

Upper airway endoscopy showed dorsal billowing of the soft palate into the pharyngeal lumen, with nasopharyngeal inflammation. During inspiration, the roof of the pharynx was partially collapsing, significantly narrowing its lumen.

In light of these findings and the inspiratory effort seen at rest, a temporary tracheostomy was performed at the junction of the upper and middle third of the neck. A 10 cm ventral midline incision was made through the skin, subcutaneous tissue and cutaneous colli muscles. The paired sternothyrohyoid muscles were bluntly divided and a transverse tracheostomy made through two adjacent cartilage rings, permitting insertion of a self-retaining metal tracheostomy tube. Venous blood analysis showed mild haemoconcentration (packed cell volume 32% and total protein 86 g/L), hyperlactataemia (2.4 mmol/L) and mild hypertriglyceridaemia (1.74 mmol/L). The gelding was started on intravenous fluid therapy (IVFT) with lactated Ringer's solution at 1900 ml/h. The horse was also administered 1.1 mg/kg intravenous (IV) flunixin (Pyroflam Norbrook, Newry, Northern Ireland) twice a day (BID) and stabilised for 24 hours before further investigation. Antimicrobial therapy was initiated empirically with intramuscular procaine penicillin (22,000 IU/kg, BID; Depocillin MSD, Walton, Milton Keynes, UK) and IV gentamicin (6.6 mg/kg, once daily [SID]; Dechra, Cheshire, Northwich, UK).

The following day, computed tomographic (CT) examination of the head showed subcutaneous gas accumulation around the rostral mandibular region and caudal aspect of the ramus of the left mandible (Figure 1). These findings were deemed to be consistent with septic sialoadenitis of the mandibular salivary gland, with possible involvement of the polystomatic sublingual salivary gland. Abscessation or rupture of the mandibular salivary gland leading to regional cellulitis was another differential considered.

Oral examination was resented by the gelding due to swelling around the masseter muscles, but brief examination showed significant swelling around the lingual frenulum

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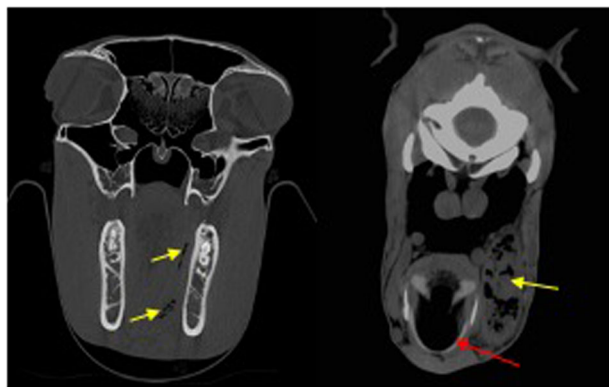


FIGURE 1 Computed tomography images at the level of the globe rostrally and at the level of the larynx caudally. Rostrally, gas accumulation (yellow arrows) was seen extending from this level to the caudal aspect of 306. In the caudal image, we see a large pocket of mixed subcutaneous gas and fluid accumulation at this level, with displacement of the larynx, pharynx, hyoid apparatus to the right side (red arrows)

and on the buccal mucosa on the left side of the oral cavity. Ultrasonography of the left mandibular region highlighted marked soft tissue swelling axial to the left hemimandible, with heterogeneity of the subcutaneous structures and anechoic fluid axial to the horizontal ramus of the left mandible, caudal to the mandibular symphysis. Abaxial to the larynx, anechoic fluid accumulation and hyperechoic structures compatible with gas/foreign bodies/bone were also visualised. Under ultrasound guidance, small stab incisions were made into the pockets of anechoic fluid rostrally at the level of the second cheek teeth on the ventral aspect of the mandible and caudally immediately axial to the junction of the vertical and horizontal rami of the left mandible. Moderate amounts of thick purulent fluid and scant necrotic soft tissue were obtained and submitted for bacterial culture and sensitivity, which yielded no growth initially. These incisions were lavaged daily with sterile saline and the gelding was changed to oral doxycycline (10 mg/kg, BID) and oral phenylbutazone (2.2 mg/kg, BID). Doxycycline was chosen on an empirical basis, as although no culture growth was obtained, this does not preclude infection due to variability of culture growth success. On Day 4, upper airway endoscopy showed marked improvement of the pharyngeal swelling and lumen diameter; therefore, the temporary tracheostomy tube was removed with no recurrence of respiratory distress.

Oral examination was repeated 7 days after admission, with the gelding restrained in stocks and sedated with 0.3 ml (6 µg/kg) detomidine (Domidine Dechra, Cheshire, Northwich, UK) and 0.5 ml (10 µg/kg) butorphanol (Dolorex MSD, Walton, Milton Keynes, UK). The swellings at the mandibular symphysis and caudal aspect of the left hemimandible persisted, showing minimal improvement with medical therapy, despite the gelding appearing brighter with an increased appetite. Oral swelling had reduced and the mandibular salivary duct opening at the left sublingual caruncle could be visualised. A 4-Fr cat urinary catheter was introduced into the duct opening and 10 ml of iohexol (Omnipaque, GE Healthcare, Marlborough, MA, USA) was injected into the catheter and a contrast sialogram obtained (Figure 2).

LEARNING POINTS/TAKE HOME MESSAGES

- Contrast sialography provided information not obtained via computed tomography and has the benefit of being achievable in general practice.
- The use of intraluminal stenting offers a viable alternative to surgery for secondary intention healing of salivary gland rupture.
- This case report shows a novel technique for achieving resolution of clinical signs associated with a sialo-cutaneous fistula without the need of general anaesthesia.

DIFFERENTIAL DIAGNOSIS

- Mandibular salivary gland rupture and sialo-cutaneous fistula;
- Sialolithiasis;
- Septic sialadenitis;
- Foreign body;
- Apical infection.

TREATMENT

The sialogram demonstrates rupture of the rostral portion of the mandibular salivary duct and sialo-cutaneous fistula. The fistula is in a distinct location separate to either of the stab incisions made on the second day of hospitalisation. Given the amount of cellulitis and secondary subcutaneous infection, surgical debridement of the fistula and primary repair of the duct laceration was not performed. It was elected to place a long-stay catheter in the duct to bypass the fistula and allow the saliva to exit through the opening in the oral cavity. The previously placed urinary catheter was removed and replaced with a 16-G over-the-wire polyurethane intravenous catheter (Mila International, Florence, KY, USA) inserted into the duct beyond the level of the fistula (Figures 3 & 4). Once inserted, the catheter was held in place with 3-metric polydioxanone (PDS, Ethicon, J&J, NJ, USA) interrupted sutures through the

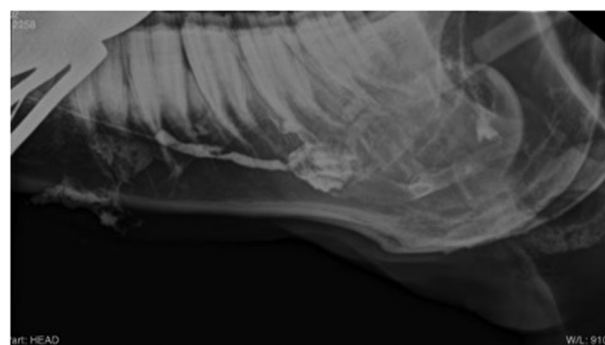


FIGURE 2 Sialography of the mandibular salivary duct indicates irregular narrowing rostrally and subsequent severe widening caudally. There are Two well-defined areas of contrast accumulation, with the most rostral one extending until the skin margin at the level of 307 (indicating a sialo-cutaneous fistula), and a Caudal emphysematous area at the level of the pharynx and larynx



FIGURE 3 Polyurethane catheter insertion into left mandibular salivary duct at the left sublingual caruncle

wings and around the hub of the catheter adjacent to the duct opening.

On Day 3 after catheterisation, the end of the catheter was displaced partially from the duct and chewed by the gelding. Repeat catheterisation was achieved under standing sedation; the sutures were replaced this time with 3-metric nylon sutures (Ethilon, Ethicon, J&J, NJ, USA) as one of the PDS sutures had broken down during the previous placement. Transcutaneous ultrasound showed improvement of the rostral swelling behind the mandibular symphysis; the swelling caudal to the left ramus of the mandible had increased in size, with a large pocket of anechoic fluid caudal to the angle of the mandible, abaxial to the laryngeal cartilages. An additional incision was made into this pocket, with a large amount of purulent fluid and caseous material obtained; this was submitted for culture and sensitivity, returning a profuse growth of *Staphylococcus aureus* resistant to multiple antimicrobial classes (Table 1) and no anaerobic growth. Antimicrobial therapy was changed to oral enrofloxacin (7.5 mg/kg, SID). The catheter was maintained in the duct for further 7 days alongside systemic antimicrobials and NSAID administration. To ensure patency of the catheter, it was flushed daily with 3 ml of saline via the oral opening with minimal resistance; lavage of the external incisions was continued as before.

TABLE 1 Aerobic culture and sensitivity for growth of *Staphylococcus aureus* isolated from mandibular abscess

Trimethoprim/sulfa	R	Penicillin	R
Oxytetracycline	R	Ampicillin	R
Doxycycline	R	Amoxycillin and clavulanate	S
Enrofloxacin	S	Ceftiofur	S
Marbofloxacin	S	Cefquinome	S
Gentamycin	R	Cefoxitin	S
Neomycin	R	Rifampicin	S
Amikacin	R	Sodium fusidate	S

R resistant, S sensitive

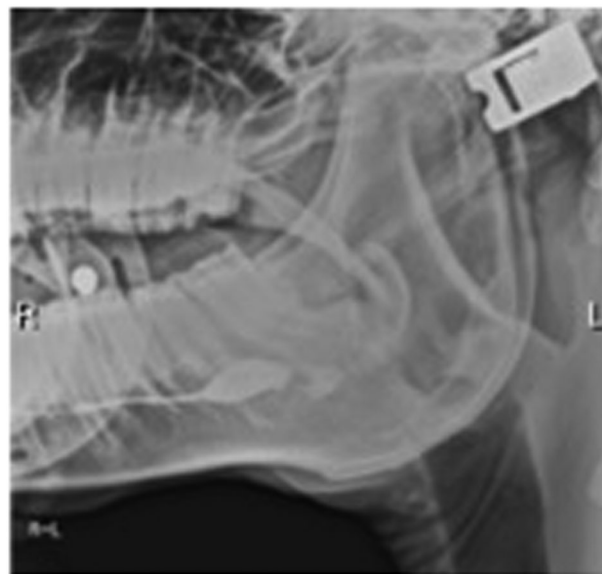


FIGURE 5 No evidence of contrast leakage from the left mandibular salivary duct at the previously described site, consistent with healing of the previously diagnosed sialo-cutaneous fistula. There is a substantial reduction in size of the ill-defined area of increased opacity containing gas at the angle of the mandible, showing improvement of the caudal region of abscessation

OUTCOME AND FOLLOW-UP

Sialography was repeated at Day 7 (Figure 5). Second intention healing of the sialo-cutaneous fistula was deemed to be achieved, as no further subcutaneous accumulation of contrast material could be visualised. The catheter was removed from the duct, no further increase in size of the swellings or increase in drainage was observed following catheter removal, and the gelding was discharged from the hospital 72 hours

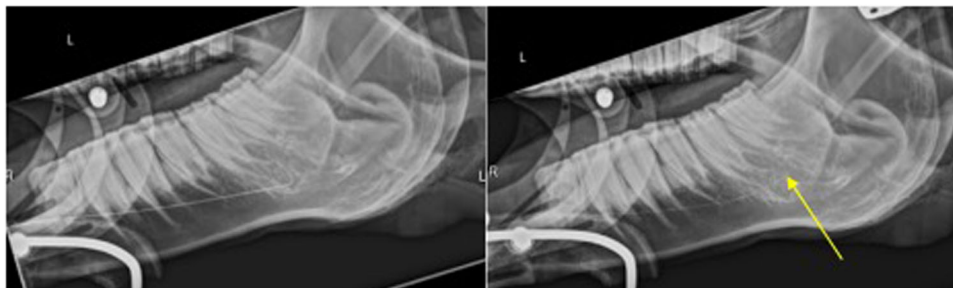


FIGURE 4 Lateral–lateral radiographs, depicting the initial placement of the guidewire through the duct opening into the salivary gland, with subsequent insertion of the radiopaque polyurethane catheter, which ends at the level of the root of 311 (yellow arrow)

later. Telephone consultation with the owner 2 months following discharge found the gelding to be doing well at home with no recurrence of swelling.

DISCUSSION

Septic sialadenitis is an uncommon condition in horses; reported in the literature as occurring secondarily to sialolithiasis.¹ Sialolithiasis could not be confirmed in this case. A small mineral opacity (8 × 3 × 3 mm) was seen on the CT examination at the lateral aspect of the oral cavity, at the level of 306. This opacity anatomically could be within the mandibular salivary duct; however, it was not encountered on catheterisation of the duct or seen on the radiographs/sialographs obtained subsequently. It seems unlikely that this opacity was the cause of the fistula, as it was too small and not within the duct, but cannot be ruled out. Other causes of sialadenitis include foreign body or dental disease causing ascending bacterial infection, which seeds into the mandibular or parotid glands. In one retrospective study, 14/15 cases had oral/dental abnormalities identified.¹ Oral examination and CT did not show any dental abnormalities, making this an unlikely cause of the sialadenitis in this case. In addition, for dental/oral abnormalities causing sialadenitis, anaerobic infection is routinely cultured (9/9 samples submitted¹). Our bacterial culture showed no anaerobic growth, growing only a multidrug-resistant *S. auerus*, which would more commonly result from commensal skin contamination or nosocomial infection.

To the authors' knowledge, this is the first report within the literature of intraluminal stenting of a salivary duct to manage salivary duct injury in a horse. This technique is widely reported within the human literature for management of Stenson's duct injury, either as a primary treatment or after surgical repair.^{2–4} Intraluminal stenting is not novel within equine surgery, as its use is advocated for maintaining the tear duct patency for lacerations involving the eyelid margins,⁵ where the catheter is maintained for 4–6 weeks following primary surgical repair.⁶ Within surgery of the oral cavity, the use of polyurethane stents has been advocated to act as a bridge between lacerated ends of the parotid duct during primary surgical repair or to protect the duct following repair.^{7,8} Primary surgical repair in this case could have resulted in a faster repair and subsequent reduction in days of hospitalisation; however, given the well-established swellings and cellulitis at the time of identification of the laceration, alongside the complication of a temporary tracheostomy and owner's constraints, an alternative approach was sought.

Catheterisation of the duct was achieved initially with a 4-Fr urinary catheter designed for use in male cats. After performing sialography and flushing the gland with sterile saline, the opening was more dilated, allowing the passage of the 16-G wire guide into the duct. The duct opening was located rostro-laterally to the base of the lingual frenulum (Figure 3). In comparison to the right side of the mouth, the left duct opening was readily identified and subsequently catheterised. This process was made easier by the enlargement of the sublingual caruncle due to chronic inflammation, similar to previous reports.⁹ Once in place, the catheter was well tolerated by the gelding and only needed to be replaced on one occa-

sion. This was caused by one of the initial sutures with polydioxanone breaking down on Day 3 and the catheter being dislodged from the duct. Once changed to nylon sutures, no further breakdown was observed and the catheter was maintained for 7 days without further complications. Suture breakdown was assumed to be accelerated due to the saliva within the oral cavity, although its use was reported without complication in the literature.⁸ Feed and water were given to the gelding ad libitum to no ill effect on the catheter placement or position. With the catheter in situ in the duct, the rostral swelling, caudal to the mandibular symphysis at the level of the fistula, rapidly improved in appearance, and within 72 hours, the contour of the rostral mandible was almost within normal limits. In our case, we aimed for 7 days of catheterisation before repeat examination based on the mean time of catheter placement following surgical removal of sialoliths in the parotid duct.⁷ This was sufficient time in this case for resolution of the fistula to occur but may need to be extended for more extensive lacerations.

The use of a polyurethane catheter in this location represents a novel technique for the management of chronic sialocutaneous fistulae and secondary sialadenitis, which alongside drainage and flushing of the abscesses allowed for complete resolution of the sialadenitis and for secondary intention healing of the fistula without the need for general anaesthesia. The procedure of catheterisation was readily performed and well tolerated by the gelding, and should be considered for cases where either the surgical site is complicated by concurrent infection or patient-level factors make general anaesthesia undesirable, or for economic reasons.

CONFLICT OF INTEREST

The authors declare they have no conflicts of interest.

ETHICS STATEMENT

No ethical approval was required for this case report. The case detailed was presented as an emergency admission, and all treatment was performed as required for the patient's welfare and with full informed consent from the owner.

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