Case Report

Computed tomography assisted surgical correction of persistent right aortic arch in a neonatal foal

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Introduction

Persistent right aortic arch (PRAA) is a congenital vascular anomaly described previously in the dog (Slatter 1993; Muldoon et al. 1997; MacPhail et al. 2001), cat (Slatter 1993), oxen (Bartels and Vaughan 1969) and horse (Bartels and Vaughan 1969; Petrick et al. 1978; Van der Linde-Sipman et al. 1979; Mackey et al. 1986; Butt et al. 1998). It occurs during embryonic development, when the right 4th aortic arch persists instead of the left. The result is the formation of a vascular ring consisting of the right aorta, pulmonary artery and ligamentum arteriosum that traps the oesophagus and trachea at the base of the heart.

Depending on the degree of constriction, only liquid is able to pass through the stenotic region. Clinical signs include failure to thrive and grow despite extreme hunger and a vigorous appetite, regurgitation of food through the mouth and nasal passages and aspiration pneumonia. In the horse, the age of onset of clinical signs varies and has been reported to range from several days of life to 14 months of age (Bartels and Vaughan 1969; Petrick et al. 1978; Van der Linde-Sipman et al. 1979; Mackey et al. 1986; Butt et al. 1998). These reports describe the condition in the horse, but only one described a case in which surgical correction of the anomaly was successful.

In this report, we describe the use of computed tomography (CT) to diagnose and precisely locate a persistent right aortic arch in a neonatal foal, thereby facilitating surgical treatment.

Case details

History

A 2-day-old Arabian colt was examined to evaluate respiratory distress since birth. The foal had been born at term and without complication, but was weak and unable to rise for the first 18 h post partum.

Clinical findings

On examination, the foal was bright and alert, although very thin and weak. The animal was tachycardic (148 beats/min) but normothermic and had a normal respiratory rate, with increased respiratory effort. Auscultation of the chest revealed increased respiratory sounds throughout both lung fields. Harsh sounds were detected on tracheal auscultation. The foal would nurse the mare vigorously, but stopped frequently due to respiratory difficulty. Milk would regurgitate from the external nares soon after suckling.

Diagnosis

Laboratory testing included haematology and blood chemistry, culture and gas analysis, a coagulation profile and immunoglobulin test (SNAP Foal Test). Abnormalities included mild hypocalcaemia (2.63 mmol/l, reference range 2.85–3.15 mmol/l), hypoproteinaemia (0.040 g/l, reference range 0.057–0.071 g/l), hypoalbuminaemia (0.018 g/l).

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Fig 1: Lateral radiograph of the thorax of the 2-day-old foal showing a patchy alveolar pattern in the ventral middle lung fields consistent with aspiration pneumonia.
reference range 0.026–0.033 g/l) and failure of passive transfer (IgG <4.0 g/l). Thoracic radiographs revealed a patchy alveolar pattern in the ventral middle lung fields, consistent with mild aspiration pneumonia. There was ventral displacement and dorsal compression of the thoracic trachea cranial to the heart, extending from the 2nd to 3rd ribspace (Figs 1 and 2). An oesophagram revealed megaoesophagus in the caudal cervical and cranial thoracic regions, the distension ending abruptly at the 2nd intercostal space (Fig 3). Ultrasonographic examination of the thorax and abdomen was within normal limits.

Based on the radiographic findings, a tentative diagnosis of persistent right aortic arch was made. The suspected vascular anomaly was further evaluated using computed tomography (GE Highlight Advantage) on Day 2 of hospitalisation. With the foal restrained under general anaesthesia and positioned in dorsal recumbency, sequential 10 mm slices were made with settings of 120 Kv, 140 mAs and a bone algorithm, centring on the cranial thorax. The images confirmed the presence of a vascular band extending from the pulmonary artery to the cranial aspect of the aorta, in the 2nd intercostal rib space (Fig 4). The aortic arch continued caudally on the right side of the heart and thorax rather than the left.

Treatment

Initial treatment consisted of potassium penicillin (22,000 u/kg bw i.v. q. 6 h), amikacin (20 mg/kg bw i.v. q. 24 h) and ranitidine (2 mg/kg bw i.v. q. 8 h). Failure of passive transfer was treated by i.v. administration of 2 l equine hyperimmune plasma. On the second day of hospitalisation, the foal was separated from the mare and given total parenteral nutrition to prevent progression of aspiration pneumonia. Surgical correction of the vascular anomaly was delayed until Day 5 to permit treatment of the pneumonia.

Surgical procedure

The foal was premedicated with midazolam and nasally intubated in a standing position to prevent regurgitation and aspiration of fluid and feed material from the dilated oesophagus (Mackey et al. 1986). General anaesthesia was induced and maintained with isofluorane gas in oxygen. A fentanyl patch (Duragesic) was applied to a shaved area in the foal’s gluteal region to provide analgesia. A small-diameter flexible nasogastric tube was inserted to the level of the obstruction to help identify the location of the oesophagus during surgery, and as a means of evaluating the diameter of the oesophagus distal to the heart base once the obstruction was relieved.

The foal was placed in right lateral recumbency and a left-lateral thoracotomy was performed through the 3rd intercostal space. Finocchietto retractors were used to spread the ribs and allow adequate access to the lesion. The left lung was retracted caudodorsally to permit inspection of the heart and greater vessels. The phrenic and vagal nerves were identified, encircled loosely with a ligature and retracted dorsally to avoid damage during the procedure. A pericardiotomy was performed to improve exposure of the greater cardiac vessels. The aorta was located in the right hemithorax, deep to the pulmonary artery. The area between the pulmonary artery and aorta was carefully bluntly dissected until the ligamentum arteriosum was exposed. It was positioned more dorsal than a comparable lesion in the dog and cat and consisted of a flat, 1 x 1 cm thick fibrous band approximately 7 cm cranial and dorsal to the heart base, running between the left pulmonary artery and right displaced aorta. The oesophagus was trapped below the ligamentum arteriosum. Movement of the nasogastric tube within the oesophagus could be detected orad to the ligament. The ligamentum arteriosum was double ligated with 0 silk on the side adjacent to the aorta, and with a single ligature on the side closest to the pulmonary artery.
Transection of the ligament between the ligatures revealed an approximately 0.3 mm lumen thought to be patent prior to transection. The soft tissues surrounding the oesophagus were bluntly dissected to permit it to expand. Despite this, when gentle pressure was applied to the nasogastric tube it was impossible to pass the constricted area. The thorax was lavaged with warm saline and a 14 Fr. chest tube was placed caudal to the 9th rib, dorsally within the thoracic cavity. The thoracotomy incision was closed in several layers. The ribs were apposed with simple interrupted sutures of #1 USP polyglyconate (Maxon) applied around the ribs cranial and caudal to the incision. The intercostal muscles were not closed primarily. The overlying muscle layers, subcutaneous tissue and skin were closed in a routine manner. Mepivacaine hydrochloride (2%) was injected caudal to the 2nd, 3rd and 4th ribs dorsal to the incision site to anaesthetise these intercostal nerves. Recovery from anaesthesia was excellent.

Post operative outcome and follow-up

A mild left forelimb lameness present for 48 h post operatively was presumed to be the result of pain associated with the thoracotomy incision. Post operative analgesia was maintained with an epidural fentanyl patch, local infiltration of mepivacaine hydrochloride along the affected ribs as required and an i.v. butorphanol drip (17.8 µg/kg bwt loading dose followed by 23.7 µg/kg bwt/h) (Sellon et al. 2001). The foal was allowed to nurse the mare for short periods (5–10 mins q. 2 h) starting 12 h after surgery and for progressively increasing periods. By 72 h post operatively the foal was nursing the mare ad libitum and total parenteral nutrition was discontinued. Antibiotic therapy was maintained for 7 days. The foal was discharged from the hospital 17 days after admission, with the recommendation not to be given access to solid food until a repeat oesophagram was performed in 6 weeks. One month after discharge, the owner reported that the foal was growing and behaving normally, with no evidence of dysphagia or coughing. A repeat oesophagram was declined as the foal appeared to be thriving. Fifty-seven days post operatively, the foal was introduced to solid food in the form of increasing small amounts of hay and grass. By post operative Day 70, the foal was eating solid food alongside the dam with no observable problems. Three months post operatively, the foal was weaned and was found dead the following day.

Necropsy did not reveal the cause of death. Evaluation of the cranial thorax showed moderate stenosis of the oesophagus at the surgical site due to encircling fibrous tissue. A mild amount of feed was impacted in the oesophagus, cranial to the lesion.

Discussion

Persistent right aortic arch (PRAA) is a developmental aberration which occurs in utero. In the normal mammalian fetus, the pericardial vasculature develops from 6 brachial arches and a pair of longitudinal dorsal aortas. The aortas fuse caudal to the level of the 4th thoracic vertebra to form the definitive descending aorta. As embryological development progresses, the first 2 arches regress. The left and right third arches form the internal carotid arteries. The right 4th arch forms the right subclavian artery while the left 4th arch forms the arterial arch. The 5th brachial arch is absent in mammals. The right 6th arch forms the pulmonary artery and the left 6th arch forms the ductus arteriosus (Noden and DeLahunta 1985).
If the 4th right aortic arch persists to form the ascending aorta instead of the left, the ductus arteriosus (which later forms the ligamentum arteriosum) becomes an encircling band that encloses the oesophagus and trachea in a vascular ring. The oesophagus is compressed against the trachea, leading to clinical signs of regurgitation, dysphagia, megaesophagus and aspiration pneumonia. Compression of the trachea can also occur; however, this is rarely sufficient to produce signs of respiratory distress.

Conditions that may produce regurgitation in a neonate include cleft palate and PRAA. Other vascular anomalies that may constrict the oesophagus, such as a double aortic arch or anomalous origin of the right subclavian and common carotid arteries, have not been reported in the horse (Bartels and Vaughan et al. 1969).

Surgical correction of a PRAA is performed by thoracotomy, with or without rib resection and subsequent transection of the ligamentum arteriosum (Petrick et al. 1978). A thoracoscopic approach has recently been described in small animals but has not been reported in the horse (MacPhail et al. 2001). Surgical correction of a PRAA has been attempted in 2 foals, through the 4th (Petrick et al. 1978) and 5th intercostal space (Mackey et al. 1986), respectively. Adequate exposure was reported in both of these cases, without the need for rib resection; however, over-retraction and rib fracture may have contributed to a difficult painful recovery in one horse (Mackey et al. 1986). In this case, radiography of the chest allowed a tentative diagnosis of a PRAA but did not reveal the precise location of the lesion. Computed tomography (CT) is a newer diagnostic tool used more frequently in small animal veterinary medicine. It is of limited use in large animals because of the diameter of the bore to the gantry, the need for a table to accommodate animals weighing more than 135 kg and the need for general anaesthesia. Also, general anaesthesia may be contraindicated in animals with respiratory difficulty and aspiration pneumonia. In this case, the foal was treated with antibiotics and total parenteral nutrition for 2 days to improve lung function, before being anaesthetised for the CT scan, which took approximately 3 mins.

The scan images indicated that the most appropriate surgical approach to the lesion was through the 3rd intercostal space. The caudal extent of the lesion could not be determined from radiographs alone, and without the availability of CT images a more caudal approach may have been made, making the surgery more difficult. While the ribs of a foal are relatively pliable, fractures have been reported in association with the use of Finochietto self-retaining retractors during thoracotomy (Mackey et al. 1986). A rib resection was unnecessary in the case reported here.

Following transection of the ligamentum arteriosum, the oesophageal tube could not be advanced past the defect, despite bougienage and extensive dissection to free the oesophagus from surrounding soft tissues. It is likely that chronic extraluminal constriction of the oesophagus may have led to some fibrosis of the oesophageal tissues. We were hopeful that the oesophagus would dilate at this site as the foal grew and through the bougienage effect of the passage of food boluses. The absence of dysphagia from the time of surgery until death supported this assumption. The foal was eating hay normally up until its death, and there was no indication of choke. At necropsy, there was a mild feed impaction orad to a region of oesophageal stenosis at the previous site of the ligamentum arteriosum. This may have resulted in problems later in life had the foal survived.

At the time of writing, surgical transection of the ligamentum arteriosum is the only viable treatment for a PRAA. In dogs, a retrospective study of 25 cases reported that 92% had excellent long-term outcomes (>6 months), although repeat oesophagography in 13 dogs showed persistent megaesophagus (Muldoon et al. 1997). Only 5 cases of PRAA have been reported in the horse. Three cases underwent surgery, of which only 1 survived >3 months; this foal was reported to be doing well 10 months post operatively. One horse died immediately post operatively of acute asphyxiation associated with purulent pneumonia. The foal in the present case died acutely; however, no cause of death was determined.

Persistent right aortic arch is a rare, congenital anomaly for which surgical correction is the treatment of choice. Although a presumptive diagnosis was made using radiography, the adjunctive use of CT more accurately diagnosed the lesion in this foal and indicated the precise location with respect to the heart and greater vessels, facilitating selection of the best surgical approach. Despite this, the prognosis for a positive long-term outcome of treatment of PRAA in horses appears poor.

Manufacturers’ addresses

1Idexx Laboratories Inc., Westbrook, Maine, USA.
2GE Medical Systems, Milwaukee, Wisconsin, USA.
3Immunogenics Inc., Ontario, New York, USA.
4Janssen Pharmaceutical Products, Titusville, New Jersey, USA.
5US Surgical, Norwalk, Connecticut, USA.

References

Clinical Commentary

Advances in diagnosis and treatment of persistent right aortic arch in a neonatal foal

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In the article above, Bauer et al. (2006) describe the diagnosis and surgical correction of persistent right aortic arch (PRAA) in a neonatal foal. This report offers at least two valuable clinical insights. The first is the utility of computed tomography (CT) for the diagnosis of intrathoracic disorders in foals. While the size of the average adult horse limits the use of CT to evaluation of the distal limb, head and more cranial portions of the cervical spine, CT can be used to examine every part of the neonatal foal, including the thorax and abdomen.

In this case, CT allowed definitive presurgical diagnosis of the anomaly when other imaging modalities had provided only a tentative diagnosis. Furthermore, CT revealed the exact location of the anomalous vessel, enabling the surgeons to select the most appropriate surgical approach. Were it not for the precise localisation of the ligamentum arteriosum preoperatively, the surgeons would probably have entered the thoracic cavity more caudally, thereby making the surgical procedure more difficult for the surgeon and more traumatic for the patient.

This case also serves as a great reminder to clinicians of what CT offers for the adult horse, despite its limited availability, expense and practical considerations (e.g. the need for the patient to be anaesthetised). The ability of CT technology to provide detailed, 3-dimensional reconstructions of the gross anatomy of a region can be a tremendous asset in both diagnosis and presurgical planning. Although the size of the average adult horse limits which portions of the body can be imaged, CT nonetheless enhances our ability to evaluate and treat various disorders of the head (e.g. oral cavity, paranasal sinuses), neck and limbs. In this way, the costs of diagnosis, treatment and lost training ultimately are minimised.

The second clinically important message in this report is that, even with prompt and accurate diagnosis, careful presurgical management, skilled surgical correction and an uncomplicated post operative recovery, foals with PRAA apparently have a poor prognosis for long-term survival. Of the 6 equine cases now reported (Bartels and Vaughan 1969; Petrick et al. 1978; Van der Linde-Sipman et al. 1979; Mackey et al. 1986; Butt et al. 1998; Bauer et al. 2006), only 1 of 4 foals that underwent surgery survived longer than 3 months post operatively.

However, it should be remembered that 4 of the 5 previous reports are at least 20 years old and perioperative care in equine neonates has advanced considerably in that time. Therefore, it could be said that this collection of cases paints too bleak a picture of PRAA in horses. In the report by Butt et al. (1998), the condition was not diagnosed until the patient was 14 months of age and evidently there are degrees of compromise or compensation associated with PRAA in horses.

Even so, the foal in this current report (Bauer et al. 2006) did not survive beyond 3 months. It had recovered uneventfully from surgery, was growing and behaving normally, and even eating solid food with no apparent problems at the time of its sudden and unexpected death at 3 months of age. Unfortunately, no cause of death was found. It seems significant that the foal was found dead the day after being weaned, which is a stressful event for any foal. As PRAA is a developmental anomaly, it is possible that the foal had another anatomical abnormality or, more likely, a functional defect not identified by routine bloodwork, CT, thoracotomy or necropsy. Van der Linde-Sipman et al. (1979) described other cardiovascular anomalies in association with PRAA in a horse, so it is entirely possible that other less obvious abnormalities of structure or function were present in the case described by Bauer et al. (2006).

Thoracoscopic correction of PRAA in a dog has recently been reported (MacPhail et al. 2001). While this surgical approach may reduce the post operative morbidity associated with surgical correction of PRAA in foals, it does not address the issue of concurrent abnormalities which may ultimately determine long-term survival in horses. Unfortunately, PRAA in horses seems to be another instance in which our ability to diagnose a condition outstrips our understanding of its aetiopathogenesis, manifestations and ramifications, and our ability to successfully treat it.
References

Persistent right aortic arch (PRAA) is a congenital anomaly in which a vestigial structure persists when it should have disappeared. In PRAA, the esophagus is constricted, primarily by a combined arterial channel that later fibroses to form the ligamentum arteriosum, resulting in esophageal dilatation and dysphagia. PRAA has been reported in small animals as well as in a calf, 3 a bull, 4 a llama, 5 a bison, 6 and foals. 7 The reports of PRAA in large animals fail to show a favorable outcome (Table 1). Various breeds can be affected. Computed tomography assisted surgical correction of persistent right aortic arch in a neonatal foal. Equine Vet Educ 2006;8(1):40-46. Scott EA, Kneller SK, Witherspoon DM.