Management of bilateral choanal atresia in a foal

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Case Description—A 1-day-old Standardbred foal with a history of extreme respiratory distress after birth consistent with upper airway obstruction was evaluated. A temporary tracheostomy tube was placed by the referring veterinarian.

Clinical Findings—On initial examination, there was evidence of hypoxic-ischemic syndrome, secondary to perinatal asphyxia. Endoscopy revealed obstruction of both nares at the level of the choanae; a diagnosis of bilateral choanal atresia was made.

Treatment and Outcome—The foal was anesthetized and underwent transendoscopic laser fenestration of the buccopharyngeal membranes. Three weeks after surgery, cicatricial narrowing of the choanae was apparent and further transendoscopic ablation was performed. Recurrent stenosis necessitated revision surgeries involving a combination of laser ablation with topical administration of mitomycin and, subsequently, a combination of radial incisions into the stenotic tissue and repeated bougienage with a cuffed endotraheal tube. The degree of stenosis decreased, and at 1 year of age, the horse was an appropriate size for its age, had choanae that were almost maximally open (> 85%), and had entered training. Mild stenosis was still evident when the horse was reexamined the following year, although there was no evidence of exercise intolerance or respiratory compromise.

Clinical Relevance—Bilateral choanal atresia in a foal can be successfully treated via transendoscopic fenestration of the buccopharyngeal membranes, enabling the horse to subsequently participate in athletic activities. Secondary problems resulting from initial asphyxia and recurrent stenosis at the surgical site can be overcome but may require prolonged and extensive treatment. (J Am Vet Med Assoc 2006;229:1784–1789)

A 1-day-old Standardbred colt foal was evaluated at the George D. Widener Hospital at the New Bolton Center, University of Pennsylvania. The foal was born the previous night; parturition had been uneventful, but immediately after birth, the foal developed extreme respiratory distress and made attempts at open-mouth breathing that were consistent with an upper respiratory tract obstruction. Within 20 to 30 minutes of birth, a temporary tracheostomy tube was placed by the referring veterinarian and the foal’s respiratory distress improved dramatically. Choanal atresia was suspected.

At the initial examination, the foal had signs of depression and was recumbent; responses to external stimuli were decreased, compared with responses expected from a clinically normal foal. Rectal temperature and pulse rate were within reference limits. The foal’s respiratory effort appeared normal but included an apneustic breathing pattern with periods of apnea of as long as 30 seconds followed by a cluster of normal deep breaths. Mucous membranes were injected and icteric, and petechiae were present; the tongue was erythematous. No meconium had been passed, and the foal had a weak suck reflex. Initial clinicopathologic analyses revealed high plasma creatinine and bilirubin concentrations, high plasma fibrinogen concentration, and marked leukopenia characterized by neutropenia with a left shift. A semiquantitative enzyme immunoassay revealed that the foal’s plasma IgG concentration was > 800 mg/dL (indicative of adequate transfer of passive immunity). An arterial blood gas analysis was performed and revealed markedly low PaO2 (43.9 mm Hg; reference range, 63 to 72 mm Hg) with 75% O2 saturation. Oxygen supplementation at a rate of 10 L/min was initiated through the tracheostomy tube; arterial blood gas analysis was repeated, and results indicated that there was improvement in blood oxygenation (PaO2, 64 mm Hg; O2 saturation, 92%). An endoscopic examination of both nares confirmed bilateral choanal atresia.

Initial procedures included placement of a clean temporary tracheostomy tube, placement of an IV catheter, and collection of blood for bacterial culture. Treatment with ticarcillin-clavulanic acid (50 mg/kg [22.7 mg/lb], IV q 6 h) was initiated. Because the foal’s changes in mentation were consistent with neonatal encephalopathy following perinatal asphyxia and it was considered unlikely that it would be able to tolerate enteral feeding for several days, IV total parenteral nutrition was provided. The foal received a mixture of dextrose (10 g/kg [4.5 g/lb]), amino acids (2 g/kg [0.9 g/lb]), and lipid emulsion (1 g/kg [0.45 g/lb]) with additional supplements of a multivitamin preparation (1 vial or 10 mL) and minerals (1 vial or 1 mL) added once during each 24-hour period. Intravenous fluid therapy (5% dextrose with 20 mEq of KCl/L, 0.61 mEq of magnesium/L [as MgSO4], 32 mEq of calcium/L [as calcium gluconate], and 3.6 mmol of phosphorus/L [as KPO4]) was administered via the IV catheter at a rate calculated to provide 4 to 5 mg of dextrose/kg/min (1.8 to 2.3 mg of dextrose/lb/min); once parenteral nutrition was established, the fluid rate was adjusted according to maintenance fluid calculations for neonates (Holliday-Segar formula). In addition, although the foal did not have signs of neonatal gastroenereopathy at this time, there were...
concerns that it may develop. The foal also received caffeine1 per rectum (10 mg/kg) as a respiratory stimulant to encourage a more normal breathing pattern.

During the next 12 hours, the foal’s respiratory pattern became more consistent and there were shorter, less frequent periods of apnea; it became more responsive to external stimuli and had an improved suckle reflex. Because the foal did not urinate voluntarily despite a palpably distended bladder, a urinary catheter was placed and a sample of urine was submitted for urinalysis and renal function assessment, including sodium fractional excretion and creatinine clearance. The foal continued to make no attempt to stand. It had signs of abdominal discomfort, including rolling onto its back and straining to defecate, which improved following administration of a soapy enema and subsequent passage of meconium. However, during the following 24 hours, the foal developed a moderate degree of abdominal distension despite passing more meconium, and ileus or neonatal gastrointestinal obstruction was suspected. Analgesia (butorphanol1 [35 µg/kg (15.9 µg/lb), IV] as required) was provided, and the foal continued to receive parenteral nutrition. During this period, the foal became increasingly more responsive and the episodes of apnea resolved. Plasma creatinine concentration decreased, and urinary fractional excretion of sodium was within reference limits; the remainder of the urinalysis was unremarkable other than trace blood, likely associated with bladder catheterization. Therefore, the foal’s condition was considered sufficiently stable for it to undergo anesthesia and correction of the choanal atresia.

On the third day after admission, the foal was anesthetized and placed in sternal recumbency. A diode laser with an 800-µm bare fiber was used transendoscopically in contact fashion to create an opening through the buccopharyngeal membrane that covered both choanal openings (Figure 1). The incision was performed in a dorsal to ventral direction through 2 layers of membranous tissue. Because of the tension on this membranous tissue, the edges retracted away from the laser fiber during transection; with minimal additional ablation and a few further incisions, a fenestration was created. The laser was set at 16 W in continuous mode, and 2,454 J was used during the procedure.

The foal sustained moderate blood loss (estimated volume, 500 mL) during surgery, and both choanae and nasal passages were packed with gauze strips. The foal recovered well from anesthesia and surgery, and although the PCV decreased mildly after surgery (from 33% immediately prior to surgery to 24% at 12 hours after surgery), the value rapidly stabilized. After cross-matching its blood type, the foal received a whole-blood transfusion (3 L) to improve its oxygen-carrying capacity. Three days after surgery, the packing was removed and the upper portion of the respiratory tract was reexamined endoscopically. Moderate airflow through the nostrils could be felt at this time. Because the respiratory tract was apparently normal, the temporary tracheostomy tube was removed, although intranasal oxygen supplementation continued; results of arterial blood gas analysis indicated that oxygenation was not adversely affected by tube removal.

During the immediate postoperative period, the foal was intermittently febrile, had harsh lung sounds bilaterally, and had a progressively increasing plasma fibrinogen concentration. Radiographic evaluation of the thorax at this time revealed a mixed alveolar and interstitial pattern consistent with pneumonia; the interstitial pattern was predominantly seen in the dorsal and dorsocaudal portions of the thorax and caudal to the cardiac silhouette with evidence of an alveolar

Figure 1—Endoscopic views of the right choana in a foal with bilateral choanal atresia before and after surgery. A—Image obtained before surgery. Notice the imperforate oropharyngeal membrane covering the choanal opening. B—Image obtained 3 days after surgery. A diode laser was used transendoscopically to create an opening through the buccopharyngeal membrane that covered the choanal openings. Pre- and postsurgical findings for the left choana were similar. Arrows show edge of ethmoid recess visible at the top of each image.
pattern within the ventral lung fields and at the cranial border of the cardiac silhouette. Pneumonia was also detected radiographically, which was assumed to be secondary to the tracheostomy. Bacterial culture of blood yielded Klebsiella oxytoca; although the organism was multidrug resistant, it had intermediate susceptibility to ticarcillin-clavulanic acid and was susceptible to cefotiofur. Treatment with cefotiofur (10 mg/kg, IV, q 6 h) was initiated, and administration of ticarcillin-clavulanic acid was discontinued. The previously described abdominal distension, and enteral feeding via an indwelling nasogastric tube was commenced. Initially, the amount fed was equivalent to 1% of body weight provided during a 24-hour period; enteral feeding was progressively increased as tolerated by the foal. Within 10 days, the foal gained weight and had learned to feed from a bottle. Once the foal’s enteral intake exceeded 10% of body weight, total parenteral nutrition was gradually discontinued during a period of 12 hours. During this 10-day period, the foal had some signs of abnormal mentation including somnolence and periods of hyperresponsiveness. These signs were considered to be consistent with neonatal encephalopathy. Thoracic radiography revealed evidence of some resolution of pneumonia, although there remained an area of potential consolidation in the caudoventral lung fields. The pneumomediastinum had resolved during the interval since tracheostomy tube removal. The temporary tracheostomy site healed without complication. Three weeks after the initial surgery, endoscopy of the upper portion of the respiratory tract revealed cicatricial narrowing of the choanal openings (Figure 2). Following sedation and local administration of an anesthetic agent, the narrowing was treated with the diode laser used transendoscopically in contact fashion (applied at 16 W; continuous mode; 3,424 J), and the foal was discharged at 6 weeks of age.

During the following 6 months, the foal was reexamined monthly at the hospital and required several revision surgeries to manage repeated stenosis of the choanae. A combination of further laser ablation of tissue and temporary stent placement was used initially. However, the stents became clogged with nasal secretions after a few days. Those procedures were later combined with topical application of mitomycin (5 mg in 15 mL [total volume]). The mitomycin was applied to each choana, over the surface of the stent, by use of a piece of soaked gauze secured externally to the stent with umbilical tape. However, the stenosis continued to recur, and an alternate technique in which radial incisions were made into the stenotic tissue with the diode laser (applied at 16 W; continuous mode; 3,000 J) followed by bougienage with a cuffed endotracheal tube (14 mm) under endoscopic guidance was applied. First, the bare diode fiber was used transendoscopically in contact fashion, and 4 to 8 radial incisions were made through the stenotic tissue from the luminal mucosal surface and extending abaxially. This was followed on successive days (typically 3 treatments) by bougienage. With the endoscope within the lumen of the endotracheal tube, the cuff was placed at the site of stricture and inflated for approximately 5 to 10 seconds to a pressure that was adequate to create moderate resi-
When the horse was 2 years old, it was returned for evaluation because of breathing-associated noise during training, although it had no signs of exercise intolerance. Endoscopic evaluation revealed mild stenosis of the choanal openings (right opening was more affected than the left). Radial incisions were again made in the stenotic tissue with the diode laser used transendoscopically, followed by bougienage on alternate days over the period of 1 week (Figure 3). Overall, the horse underwent 12 transendoscopic laser procedures during a period of 2 years; the treatments enabled the horse to undergo race training without evidence of respiratory compromise.

**Discussion**

Choanal atresia is a relatively rare congenital malformation, and affected individuals lack communication between the nasal cavity and the pharynx. Multiple theories have been proposed for its embryologic origin in humans including failure of the bucchopharyngeal membrane to rupture during early gestation, persistence of the nasobuccal membrane of Hochstetter, abnormal persistence or location of mesodermal adhesions in the nasal choanal region, and misdirection of mesodermal flow caused by errors in neural crest migration during early nasal and facial development. This last theory is supported by evidence (confirmed by use of CT analysis) of structural alteration of several craniofacial bones in humans with choanal atresia. These alterations include medial displacement of the pterygoid plates and lateral thickening of the vomer, narrowing of the nasal cavity, malformation of the skull base including absence of the cribiform plate, and patent foramen cecum with nasal dermoid cyst and sinus. Choanal atresia has also been detected in humans with rare craniofacial clefts, which may also result from abnormal neural crest cell migration.

In addition to humans and horses, choanal atresia has also been identified in several other species, including llamas, sheep, dogs, gorillas, and baboons. Atresia may be unilateral or bilateral and composed of bony, membranous, or mixed bony-membranous tissues. Human congenital choanal atresia was first described in 1755 by Roeder, and it occurs in approximately 0.82 of 10,000 births depending on birth location, according to analysis of birth registries of congenital anomalies. Complete bony atresia (29%) or mixed bony-membranous anomalies (71%) were detected most commonly when CT scans were analyzed. Unilateral and bilateral choanal atresia has also been described in horses, but the true incidence and etiology in that species have not been elucidated. Accuracy of estimations of the true incidence in horses may be affected by the life-threatening nature of the airway obstruction in bilaterally affected foals, which can be fatal unless parturition is closely attended and an alternative airway is provided to these obligate nose-breathers.

In horses, the tissue that obstructs the choanal opening is typically described as membranous, as it was in the foal of this report. However, to our knowledge, results of additional diagnostic procedures (eg, CT) to confirm this have not been reported. Computed tomographic findings in an affected llama cria have been described, and CT is used extensively in humans to characterize the nature of the atretic tissue and assess patients for other craniofacial and nasal anomalies. Narrowing of the caudal portion of the ventral meatus on endoscopic examination has been suggested in a previous report in a horse and narrowing of the nasal passages and nasopharynx is well described in humans with choanal atresia. In horses, adjunctive imaging modalities, such as CT or magnetic resonance imaging, may be useful to qualitatively determine whether the composition of the atretic tissue is membranous as previously suggested, and evaluate affected individuals for evidence of other craniofacial anomalies.

Familial cases of choanal atresia in humans have been reported. Many of those children had consanguineous parents, suggesting an autosomal recessive trait; however, more commonly, choanal atresia occurs sporadically in human populations and any genetic influence is likely multifactorial. On the basis of evaluations of alpacas and llamas with unilateral and bilateral choanal atresia, a hereditary link (with clusters of affected crias born to the same or closely related dams) has been suggested; however, the mode of inheritance in those cases appears more complex than a simple autosomal recessive trait. Although several reports of choanal atresia in horses have involved Thoroughbreds, a larger case population is needed to make a meaningful assessment of heredity.

Administration of drugs that inhibit the synthesis of thyroid gland hormones (specifically methimazole and carbimazole) to humans during early gestation has been associated with choanal atresia in their offspring. Choanal atresia is also reported to occur more frequently among children born to mothers who were exposed to drinking water contaminants (specifically chlorinated solvents such as trichloroethylene) during gestation than it does among children born to unexposed mothers. No obvious toxicologic cause of the condition was identified in the foal of this report.

Previously described methods for treatment of choanal atresia in horses include transendoscopic ablation of the tissue by use of a neodymium:yttrium aluminum garnet laser and a bilateral paramesial sinus flap approach to gain access to the obstructed choanae and allow resection of obstructing tissue. However, to our knowledge, there are no reports of long-term outcome or ability to return to athletic activity after either treatment. Although the paramesial sinus flap approach afforded unobstructed access to the atretic plate and caudal nasal septum in the foal of 1 report, disruption of facial suture lines resulted in facial deformity involving dental malocclusion that was attributable to asynchronism of mandibular and maxillary growth. That horse was later euthanized because of severe pneumonia. Laser ablation (performed with endoscopic guidance) of the obstructing tissue was substantially less invasive, but follow-up on that unilaterally affected foal suggested a reduction in airflow and excessive respiratory noise during exercise, which was consistent with choanal stenosis, although further endoscopic evaluation was not performed.

Although a variety of procedures for the management of bilateral choanal atresia have been described in the human medical literature, including transpalatal...
and endoscope- or microscope-guided transnasal approaches (including sharp or laser techniques), anatomic differences along with differences in the composition of the atretic plate make application of those techniques to horses difficult. Similar to findings in llamas, most affected humans have a bony or mixed bony-membranous obstruction, whereas membranous tissue is frequently present in horses with choanal atresia.

The high rate of posttreatment recurrence of stenosis that is sufficiently severe to require additional intervention (mean number of surgeries required for a successful outcome in humans with bilateral choanal atresia is 4.9) has prompted the development of multiple adjunctive therapies and techniques in human medicine. Stents are frequently used after surgery and reduce the risk of recurrence of stenosis in some people. However, inappropriately large stents may induce pressure necrosis and cause discomfort. The optimal duration for stent placement remains controversial with recommendations varying from 24 to 48 hours to 18 weeks. In foals, including the foal of this report, stents quickly become plugged with nasal secretions and their patency is difficult to maintain over the period that is required to establish an adequate airway. In addition, the size of the nasal passage limits the diameter of stent that can be inserted, and consequently, the selected stent is markedly smaller than the potential choanal opening. This limits establishment of an airway of maximal diameter in choanal atresia–affected horses.

Techniques to preserve the soft tissues surrounding the choanae by creating mucosal flaps prior to removing the bony atretic plate have also been described. Inclusion of these procedures in the surgical treatment of humans with choanal atresia resulted in less scarring at the surgical site and a reduced risk of stenosis. Development of synechiae and stenosis is less likely if circumferential mucosal stripping or injury is avoided. The positive results achieved by use of radial incisions and repeated bougienage in the management of recurrent choanal stenosis in the foal of this report support this finding. Earlier application of this technique in the management of the foal may have substantially reduced the number of surgeries required to complete treatment (of the 12 laser surgeries that the foal underwent, radial incision-bougienage procedures represented 4 of the treatments). Balloon dilatation has also been used as the sole treatment of stenosis following primary choanal repair. In several studies of humans with choanal atresia, increased success rates, decreased scar formation, reduced requirement for revision surgery, and avoidance of postoperative stenting have been achieved by use of topical treatment of the surgical site with mitomycin following completion of traditional surgical repair. Topical mitomycin treatment reduces scar formation at the surgery site through its antifibroblast activity. However, in the foal of this report, no appreciable effect of topically administered mitomycin on the degree of fibrosis and stenosis that developed at the surgery site was detected. This may be a result of the difficulty we had in maintaining direct contact between the drug and surgery site; slow-release and injectable forms of mitomycin may be more beneficial.

This case report highlights that bilateral choanal atresia in a foal can be successfully treated, enabling the horse to participate in athletic activities. Secondary problems resulting from initial asphyxia and recurrent stenosis at the surgical site should be anticipated and may be associated with prolonged and extensive treatment. In addition, rapid recognition of the airway obstruction and appropriate management immediately after birth are crucial for survival of an affected foal. Transendoscopic surgery is effective in perforating the buccopharyngeal membrane, and procedures involving radial incisions with bougienage are effective in the management of recurrent choanal stenosis and should be considered as an alternative to stenting or repeated circumferential resection of stenotic tissue.

References