

DOI 10.26873/SVR-1832-2024 UDC 591.432:599.723.12:616-072.1:616-089.8:616.24-002

Pages: 299-305

Endoscopic and Surgical Intervention of Complete Esophageal Stricture in a One-month-old Thoroughbred Foal With Rhodococcus equipneumonia

Key words

balloon dilation; foal; esophageal stricture; esophagostomy; pneumonia

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Abstract: This case report describes a surgical and endoscopic approach for treating a complete esophageal stricture in a one-month-old Thoroughbred foal with *Rhodococcus* equi pneumonia. Clinical symptoms included coughing, nasal discharge, regurgitation, and lethargy. Endoscopic and ultrasonographic examinations revealed a complete esophageal obstruction caused by soft tissue stricture surrounding the esophageal lumen. The tube esophagostomy, combined anterograde retrograde esophageal assessment, and endoscopic balloon dilation were sequentially applied for the stricture lesion, along with the critical care for the secondary complications. After treatment, the foal was allowed to eat ground feed, chopped hay, and milk, and showed improved vitality. The foal was discharged on post-admission day 35.

Introduction

Esophageal stricture (ES) is a common disorder in horses usually caused by external or internal trauma of the esophageal wall leading to narrowing of the lumen (1, 2). The primary clinical sign is deglutition related to fibrosis and scar tissue formation (2). The impaired passage in the esophagus is manifested by ptyalism, coughing, dysphagia, and regurgitation with complications including aspiration pneumonia, esophageal diverticula, and inflammation (1, 3) . Three types of strictures are recognized, affecting the types of treatment and prognosis (1, 4) : type I, lesions involving the tunica muscularis and adventitia; type II, cicatrix or fibrous rings involving the mucosa and submucosa; type III, strictures involving all layers of the esophageal wall.

Along with the physical examination, several diagnostic tools, such as endoscopy, ultrasonography, and radiography, can be used for esophageal evaluation (4, 5). Surgical interventions and alternative therapies, such as balloon

dilation, have been applied for existing strictures (1,6). The use of balloon dilation is most recently described in equine medicine, and it offers relatively safe and successful outcomes without complications in other methods (2,5,7). On the other hand, the overall prognosis of ES is guarded, especially in foals less than one year of age, because of the high rate of recurrence and concurrent esophageal diseases (3,6,7).

Pneumonia caused by the intracellular bacterium $Rhodococcus\ equi\ (R.\ equi)$ is a significant cause of disease and death in foals (8,9). The distribution of $R.\ equi$ is worldwide, and infection occurs by the inhalation of dust contaminated with virulent $R.\ equi\ (8,9)$. It is unknown why some foals suffer $R.\ equi$ infection with or without clinical findings, while some do not (10-12). According to the epidemiological data in the US, clinical finding are observed in 10-20% of foals in endemic farms, but ultrasonographic evidence of

lung abscessation have been reported to vary between 30% and 60% (10-13). Moreover, extrapulmonary disorders are not such rare as was believed (10). Immunocompromised hosts are highly susceptible to R. equi, leading to opportunistic and complicated pneumonia with clinical signs, such as cough, fever, lethargy, and increased effort and rate of respiration (8).

This paper reports a case of complete ES in a one-monthold Thoroughbred foal with R. equi pneumonia. A tube esophagostomy was performed before balloon dilation 1) to use combined anterograde and retrograde esophageal endoscopy to assess the stricture lesion from both sides, 2) to use tube esophagostomy for enteral nutritional support, and 3) to make time for endoscopic balloon wire preparation. This article describes the sequential therapies and critical care process as well as the limitations of the case, expanding the current knowledge for ES and related considerations in horses.

Case Presentation

A one-month-old, 95 kg male Thoroughbred foal was referred for the clinical signs of coughing, nasal discharge, phlegm, regurgitation, and lethargy. According to the owner and referring veterinarian, the respiratory symptoms were recognized more than 10 days before presentation and became gradually aggravated despite receiving symptomatic treatment, including a nonsteroidal anti-inflammatory drug (NSAID; flunixin meglumine, 1.1 mg/kg, IV) and antibiotics (procaine penicillin G, 22,000 IU/kg, IM).

A physical examination of the horse revealed a heart rate of 84 beats/min, a body temperature of 38.8 °C, and a respiratory rate of 60 breaths/min with green-milky nasal discharge, mild depression, and bilateral crackling sound on lung auscultation. In the laboratory analysis, neutrophilia (12.47 × 10 9 /L; reference range 2.3-9.5 × 10 9 /L), hyperglycemia (114 mg/dL; reference range 65-110 mg/dL), hypocalcemia (10.8 mg/dL; reference range 11.5-14.2 mg/ dL), elevation of total bilirubin (2.3 mg/dL; reference range, 0.5-2.3 mg/dL) and gamma-glutamyl transferase (35 U/L; reference range, 5-24 U/L), and insufficient passive immune transfer (IgG < 400 mg/dL; reference range, > 800 mg/dL).

On diagnostic endoscopy, a yellowish-white fluid, presumably a milky mixture, was visible in the upper respiratory tract and trachea (Figure 1A). Further esophagoscopy revealed a complete esophageal obstruction caused by soft tissue stricture surrounding the lumen (Figure 1B). Cervical ultrasonography confirmed the location and size of the stricture, which was located at the level of the 3rd cervical vertebra with a diameter of 1.1 cm and length of 2 cm (Figures 1C, D). The normal cervical esophagus is not visible on plain radiographs; however, using negative-contrast radiography with air insufflation, we could identify the esophagus. The

absence of air shadows beyond the ES lesion confirmed a complete obstruction (Figure 1E). In ultrasonography and contrast radiography, there was no change in the diameter of the esophagus, and the most significant changes were observed in the mucosa and submucosa, while the adventitia remained intact (Figure 1C, D, E). Based on these findings, it was inferred to be a type II stricture. In thoracic radiography, the outline of the caudal vena cava and caudal silhouette of the heart were obscured by the caudoventral opacity, and multiple patchy opacities were observed in the entire lung (Figure 4A). Thoracic ultrasonography showed roughening of the pleural surface with multiple comet-tail artifacts. The subsequent laboratory microbiology analysis using trans-tracheal wash (TTW) fluid revealed a positive R. equi result among nine respiratory disease-causing agents (IDEXX Reference Laboratories, USA).

Based on the physical examination and imaging, complete ES with R. equi pneumonia was diagnosed. Owing to the difficulty of anterograde passage and feeding, tube esophagostomy was performed distal to the stricture to bypass the lesion for enteral nutritional supply and approach the stricture retrogradely. Under sedation and intravenous anesthesia (diazepam 0.1 mg/kg and ketamine 2.2 mg/kg), the foal was positioned in right lateral recumbency, and the surgical site was prepared aseptically. The flexible endoscope was first inserted anterogradely as a guide; the incision site was chosen around the level of the 3rd cervical vertebra. A linear skin incision, approximately 10 cm in length, was made ventral and parallel to the left jugular vein. The brachiocephalic and sternocephalic muscles were separated bluntly, and the carotid sheath was retracted to expose the esophagus. A 4 cm long longitudinal incision, approximately 3 cm distal to the stricture, was made through the esophagus using endoscopic transillumination as a guide for finding the lesion and incision site (Figures 2A, B). An endoscopy through an esophageal incision, the stricture lesion, and the remaining region distal to the stricture of the esophagus as well as the stomach were assessed endoscopically. After confirming no abnormalities except for the stricture site, the esophagostomy tube was inserted and secured to the skin with sutures and bandages (Figure 2C).

During the hospitalized period, the foal was treated with a nonsteroidal anti-inflammatory drug (flunixin meglumine, 1.1 mg/kg, IV, q12hr), antibiotics (ceftiofur, 2.2 mg/kg, IV, q24hr; azithromycin, 10 mg/kg, PO, q24hr; rifampin, 5 mg/ kg, PO, q12hr), gastroprotectants (omeprazole, 4 mg/kg, PO, q24hr; ranitidine 6.6 mg/kg, IV, q8hr), and nebulization (fluticasone 2mg/kg, 10% acetylcysteine 3ml, gentamicin 2mg/kg, q12hr) with oxygen. For nutritional support, the foal received fluid (Hartmann's solution) and parenteral nutrition (OLIMEL® N9E, Baxter) under glucose monitoring until a tube esophagostomy was applied for nutritional supply. Because of the limited amount of mare's milk, commercial powdered goat's milk was supplied via the tube esophagostomy route. After the installation of an esophagostomy tube, feeding with milk was initiated at 1% of body weight

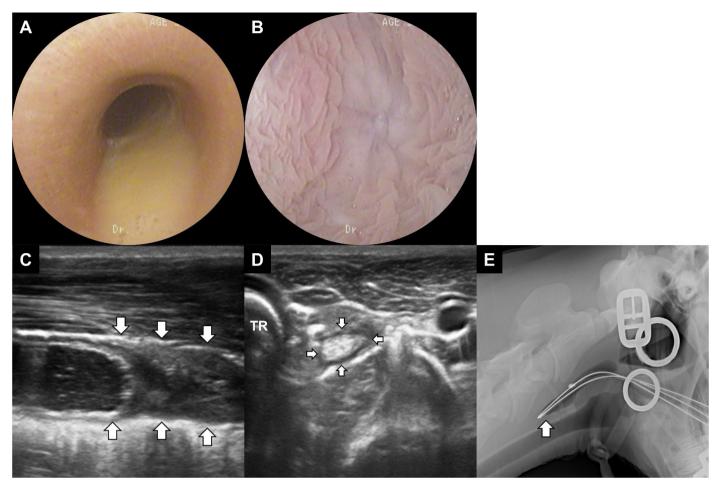


Figure 1: Endoscopic (A, B), ultrasonographic (C, D), and radiographic (E) images of the stricture lesion (white arrows) in the esophagus. (A) The trachea is filled with milky fluid. (B) The esophagus is entirely obstructed by the soft tissue stricture. (C, D) The longitudinal and transverse view of the stricture lesion on ultrasonography. TR indicates trachea. (E) Negative-contrast radiography through air insufflation was performed, confirming complete esophageal obstruction

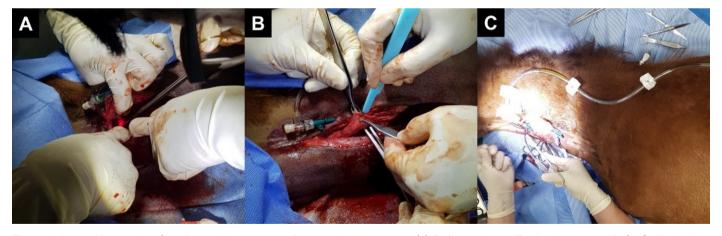


Figure 2: Surgical intervention for tube esophagostomy and retrograde assessment. (A) Endoscopic transillumination as a guide for finding lesions and the incision site. (B) Incision on the esophagus, approximately 3 cm distal to the stricture. (C) Esophagostomy tube application and securement for nutritional support

per day, and gradually increased over 10 days to 10% of body weight per day.

The complications during the treatment and enteral supply were diarrhea, fever, and weight loss. Despite the nutritional support, the foal lost 10% of body weight during the first

week, but the diarrhea improved gradually, and the body weight soon increased slowly.

Seven days after presentation, endoscopic balloon dilation was performed using a controlled radial expansion wireguided balloon catheter (CRE wire-guided dilator, Boston

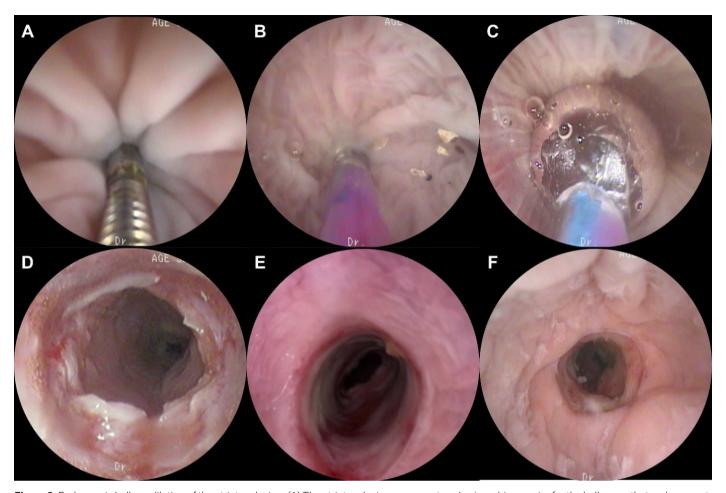


Figure 3: Endoscopic balloon dilation of the stricture lesion. (A) The stricture lesion was punctured using a biopsy wire for the balloon catheter placement. (B) The balloon catheter was placed into the stricture. (C) The balloon was inflated to reach the desired diameter stated by the manufacturer. (D) Widened esophageal lumen with mild bleeding immediately after the balloon dilation. (E) The stricture area six days post balloon dilation. (F) The stricture area 26 days post balloon dilation

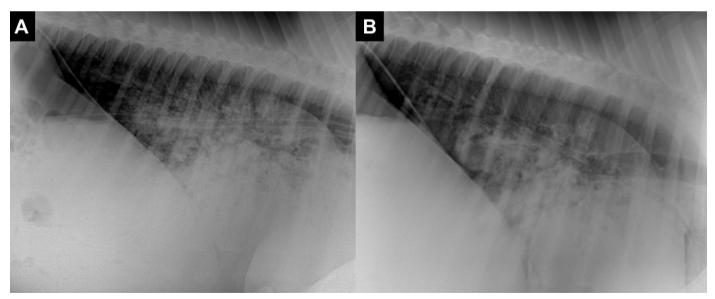


Figure 4: The radiographic image on pre- (A) and post-treatment (B) of the lung in this study. (A) The outline of the caudal vena cava and caudal silhouette of the heart were obscured due to the caudoventral opacity, and multiple patchy opacities were observed in the entire lung. (B) The patchy opacities in the lung and silhouette of the heart improved significantly after treatment (30 days post admission)

Scientific. Natick, MA, USA). The appropriate size of the balloon dilator was prepared based on the measured diameter and length of the stricture lesion. Initially, the stricture

was punctured using a biopsy wire under visualization of the lesion with anterograde and retrograde esophagoscopy (Figure 3A). Subsequently, the balloon catheter was placed into the stricture and inflated to reach the desired diameter according to the manufacturer's instructions (Figures 3B, C). The pressure and diameter of the balloon were maintained for 90 seconds with the high-pressure inflation device designed for balloon inflation and deflation (Encore 26 Inflator, Boston Scientific. Natick, MA, USA). After the procedure, the mucosa of the esophagus was closely evaluated (Figure 3D), and topical steroid injection (Triamcinolone, 1mg/site, total 6mg) into the submucosa was performed trans-endoscopically. The esophagostomy tube was removed, and a nasogastric tube was placed and secured for re-epithelialization and nutrition supply. The incision site of the tube esophagostomy was restored well without complications.

Six days after the first balloon dilation, the nasogastric feeding tube was removed, and the stricture lesion was assessed. The diameter of the stricture lesion was smaller than the adjacent normal tissue, and the scar tissue remained (Figure 3E). Subsequently, the foal was permitted to consume ground feed and chopped hay, in addition to commercial goat's milk formula, due to occasional coughing or regurgitation observed after the foal consumed a mass of grass. This dietary management was continued even after the foal was discharged. The second and third balloon dilation was also performed one week apart to widen the lesion, but it did not greatly affect the diameter. The diameter of the dilated lesion was maintained until discharge on post-admission day 35 (Figure 3F). The obscured heart silhouette and multiple patchy opacities in the lung radiography caused by aspiration and R. equi pneumonia were also improved (Figure 4B). On the other hand, in the long-term follow-up, the foal showed colic signs one month after discharge. The owner decided against further treatment and the foal was euthanized without referral for treatment or post-mortem necropsy.

Discussion

The primary causes of ES in equines include esophageal obstruction, trauma, nasogastric intubation, congenital defects, or complications following esophageal surgery (4, 6, 7). Strictures occur due to the formation of fibrous tissue and the deposition of collagen, which are stimulated by esophageal lesions (4, 6, 7). In human studies, peptic stenosis is identified as the most common cause of esophageal strictures, accounting for 70-75% of cases, resulting from gastric acid exposure (14-16). Other causes include the ingestion of caustic products, radiotherapy, foreign bodies, and infections (14-16). In this report, the precise cause of the ES is challenging to determine. However, considering that the lesion extensively covers a specific area of the esophagus and has progressed to complete obstruction, it is presumed that the lesion-potentially caused by trauma, impaction, peptic ulcer, or other factors-has developed into stenosis over a long period. Moreover, considering the horse's young age, the presence of a congenital lesion cannot be ruled out.

Despite the common occurrence of ES in horses and the several reports of successful applications of balloon dilation in stricture lesion in partial obstruction (5, 7, 17), there are no reports on complete ES in equine cases. In humans, combined anterograde retrograde esophageal dilation (CARD) was used for complete ES through the gastrostomy tube and flexible endoscope (18, 19). Horses are suitable for applying CARD because the length of the equine neck is much longer than a human and the retrograde approach through esophagostomy is more straightforward than that through a gastrostomy. Therefore, CARD was applied in this case, and esophagostomy was used for the retrograde approach and nutritional support. This is the first case report regarding the application of CARD in equine species describing the sequence of surgical intervention and endoscopic dilation, post-operative care, and complications.

In general, the prognosis of ES in foals is poor (7, 20). The stricture and concurrent complications, including megaesophagus, aspiration pneumonia, and stricture recurrence, adversely affect the prognosis (4, 7, 20). In this study, the foal had R. equi pneumonia with insufficient passive immune transfer. The pneumonia caused by R. equi infection in Korea is assumed to be prevalent (21), aggravating the disease in immunocompromised foals. When the foals have symptoms of respiratory disease, the practitioners and owners often administer drugs targeting R. equi without conducting a thorough examination, leading to a delayed diagnosis of ES. In particular, foals less than one month old typically ingest mare's milk rather than solid food, and the symptoms caused by ES are not evident until the route is completely obstructed. The similarities of the respiratory signs caused by ES and an R. equi infection might result in a misdiagnosis and delay suitable treatment. Consequently, it is suggested that the practitioners include ES in a differential diagnosis of respiratory disease in foals for the early detection of ES to achieve a better prognosis. In addition, large-scale investigations regarding the R. equi epidemiology in Korea are warranted for managing the current situation of R. equi infections, which have never been conducted.

While it is difficult to attribute the impact of *R. equi* pneumonia directly on ES, it is evident that *R. equi* combined with complications such as aspiration pneumonia, nutritional deficiency, and stress caused by ES, worsened the patient's prognosis, highlighting the importance of critical care. To meet the primary nutritional needs resulting from complete esophageal obstruction, an esophagostomy was performed initially to create a bypass for nutrient delivery. Additionally, to prevent aspiration pneumonia and secondary infections commonly associated with ES, broadspectrum, third-generation cephalosporin (ceftiofur) was administered. The pneumonia caused by biofilm-forming bacteria *R. equi* can exacerbate respiratory symptoms and

increase mortality rates in foals, so a combination of antibiotics (azithromycin, rifampin) targeting these pathogens was used. Considering that stimulation by gastric acid is a major contributing factor to ES (15, 16), antacids, including a proton pump inhibitor (omeprazole) and an H2 blocker (ranitidine), were administered throughout the period of hospitalization. Addressing pneumonia alongside ES is crucial for improving the survival rates of young foals, leading to the administration of nebulization containing antibiotics (gentamicin), steroid (fluticasone), and mucolytic agent (acetylcysteine) as well as intravenous NSAID (flunixin meglumine). Although TTW results did not identify significant pathogenic bacteria other than R. equi, a broad spectrum of medications was deemed necessary to counteract the potential lethality of infections and inflammations in immunocompromised young foal in this case. The horse's condition was closely monitored, with regular blood tests to detect any potential side effects.

Dilation of the esophagus is a relatively high-risk procedure, and there are no guidelines for clinicians in veterinary medicine on how to perform the practice safely. Although the authors referred to the guidelines for humans and previous animal case studies (4, 5, 7, 17, 22, 23), the scarcity of resources available for complete ES in horses presents several limitations for optimal care, particularly for young foals. First, there is no grading system and treatment protocols to assess the condition of the ES beneficial for selecting a proper treatment to deal with the emergency. The degree of severity and progression could not be rated in the present case and consequently there were limitations to the process of the treatments for a favorable prognosis. Second, an in-depth examination, such as histopathology and cross-sectional imaging (CT), could not be included during the procedure to exclude malignancies, as in human protocols, because of the cost and lack of resources (22). Lastly, the lack of experience and knowledge by veterinarians and owners can result in a delayed diagnosis and mistreatment. Thus, regular education and system establishment for suitable care are necessary for the horse industry.

Conclusions

In conclusion, this is the first case report of complete ES in a one-month-old Thoroughbred foal with *R. equi* pneumonia. This paper described the sequential therapies and critical care process as well as the limitations of the case, expanding the current knowledge of ES and the related considerations in horses. The data will benefit practitioners and owners by contributing to a better understanding of ES in the foals.

Acknowledgments

The authors thank all KRA staffs who participated in this study.

Conflict of Interest. The authors declare no conflicts of interest...

Author contributions. Conceptualization: J Yoon, and T Park; Data curation: J Yoon, A Kim, YB Kwak, and J Lee; Formal analysis: J Yoon; Resources: A Kim, and J Lee; Supervision: I-S Choi, and T Park; Validation: J Yoon, I-S Choi, and T Park; Visualization: J Yoon; Writing-original draft: J Yoon; Writing-review & editing: J Yoon, YB Kwak, and T Park.

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Endoskopska in kirurška obravnava popolne strikture požiralnika pri enomesečnem čistokrvnem žrebetu s pljučnico, povzročeno z *Rhodococcus equi*

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Izvleček: V poročilu o primeru je opisana kirurška in endoskopska obravnava popolne strikture požiralnika pri enomesečnem čistokrvnem žrebetu s pljučnico, povzročeno z *Rhodococcus equi (Rhodococcus equi pneumonia)*. Klinični simptomi so bili kašelj, izcedek iz nosu, regurgitacija in letargija. Endoskopska in ultrazvočna preiskava je pokazala popolno obstrukcijo požiralnika, ki jo je povzročila striktura mehkih tkiv okoli lumna požiralnika. Za zdravljenje strikture smo zaporedno izvedli cevno ezofagostomo, kombinirano anterogradno retrogradno oceno požiralnika in endoskopsko balonsko dilatacijo skupaj s kritično oskrbo sekundarnih zapletov. Po zdravljenju je žrebe lahko jedlo mleto krmo, rezano seno in mleko, njegova vitalnost pa se je izboljšala. Žrebe je bilo odpuščeno 35. dan po sprejemu.

Ključne besede: balonska dilatacija; žrebe; ezofagealna striktura; ezofagostoma; pljučnica