Magnetic compression stricturoplasty in patients with severe stricture after simultaneous esophageal atresia and duodenal obstruction repair: A case report

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Abstract. Combined esophageal atresia (EA), tracheoesophageal fistula (TEF) and duodenal obstruction result in various challenges in management, and a well-defined management protocol is still lacking. Esophageal stricture is the most common complication after EA repair. The use of magnetic compression alimentary tract anastomosis has been reported in children. By searching the literature, the present study reports the first case of simultaneous repair (EA repair followed by duodenal obstruction repair) and magnetic compression stricturoplasty for refractory esophageal stricture after EA repair in two male neonates. One of the neonates received delayed treatment of duodenal obstruction, and the other successfully underwent a simultaneous emergency operation of these combined anomalies. These two infants developed refractory strictures despite multiple endoscopic dilatation procedures during the postoperative follow-up period. Magnetic compression stricturoplasty procedures were successfully performed under fluoroscopic and endoscopic guidance without any leakage or complication. At the follow-up 10-months after stricturoplasty, the two patients achieved durable esophageal patency in the absence of dysphagia. Combination of early chest and abdominal X-ray detection is recommended to avoid a delayed diagnosis and treatment, as well as the synchronous operation for EA/TEF repair and duodenoduodenostomy in a single surgery for combined EA/TEF and duodenal obstructions. Therefore, magnetic compression stricturoplasty is a feasible and efficient method for establishing early patency of the esophagus in patients with refractory EA stricture.

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Introduction

Combined esophageal atresia/tracheoesophageal fistula (EA/TEF) and duodenal obstruction is very rare, and is accompanied with high morbidity and mortality rates (1-3). The combined anomalies always pose challenges for disease management. At present, management protocols for these combined anomalies have not yet been clearly defined, and it remains a controversial issue whether the anomalies occur together or are staged (3-6). Esophageal anastomotic stricture is a frequent complication following EA repair (7). In some cases, stricture may be recalcitrant in spite of dilatation procedures (8). For these patients, a variety of treatment methods have been tried and employed, including steroids (9), mitomycin C (10), bougie and balloon dilatation, and esophageal stenting (11,12), with varying efficiencies and success rates. However, these conventional treatment options are not useful in treating recurring hypertrophic scar tissue, and some of these patients require repeated thoracotomy with segmental esophageal resection and reanastomosis or esophageal replacement (13-14). Therefore, there is an urgent need for a treatment option for esophageal stricture in children that can effectively remove the hyperplastic scar tissues formed in the esophagus without causing trauma. In the last decade, magnetic compression anastomosis has been used to treat anastomotic stenosis following esophagoesophagostomy for EA (15-17).

The present case report describes our first experience with simultaneous repair of a combination of gastrointestinal anomalies and magnetic compression stricturoplasty treating refractory stricture after EA repair in two male neonates.

The clinical experience and data concerning the diagnosis and treatment of complex multiple digestive tract obstructions in infants with combined EA/TEF and severe duodenal obstruction were recorded and analyzed. Moreover, important properties of magnetic compression stricturoplasty while treating refractory stricture following EA repair are discussed.

Case report

Patient 1. The first case was a 2 years and 2 months old male infant, who was born at 38 weeks of gestation via cesarean section due to a uterine scar and premature rupture of the membranes, with the umbilical cord wrapping around the neck by 360˚. An excessive amount of amniotic fluid was noted on a prenatal ultrasound. His Apgar score (14) were 10 at 1 min, 10 at 5 min, and 10 at 10 min. Chest X-rays showed that the nasogastric tube (NGT) was located in the upper esophagus, suggesting EA (Fig. 1A). On the subsequent day after birth, the patient underwent the first urgent surgery, comprising right posterolateral thoracotomy and extrapleural separation for EA with proximal TEF (Gross type C) and malformation repair. On day 16 after the first operation, repeated upper gastrography verified patency of the esophageal anastomosis and a delayed diagnosis of duodenal obstruction (Fig. 1B). The patient therefore underwent an emergency exploratory laparotomy and duodenoduodenostomy for the duodenal obstruction repair. On day 4 after the second operation, enteral feeding was started after a swallow test. The infant was able to take full volume oral feeding by day 12, indicating the absence of esophageal or duodenal leakage. The infant was in good health at the 14-month follow-up. Over the next 12 months, the narrowing progressed to a refractory stricture in spite of multiple endoscopic dilatation procedures. On the 4th attempt at wire-guided balloon dilatation, it was observed that the stricture had essentially caused a severe (nearly complete) esophageal obstruction (<3 mm; Fig. 1C). As a result, the patient was a candidate for stent placement or segmental resection/anastomosis. A detailed discussion on stent placement or thoracotomy with attempted segmental resection and anastomosis was conducted with the parents of the patient, who refused the procedure due to the risk of complications such as restenosis. Ultimately, according to medical opinion, the patient received an endoscopy-guided magnetic esophageal compression stricturoplasty.

Patient 2. The second case was another 1 years and 11 months old male infant who was born at 39 weeks of gestation through vaginal delivery, weighing 2,540 g. The Apgar scores were 8, 9 and 9 at 1, 5 and 10 min after birth, respectively. The prenatal ultrasound showed polyhydramnios. Spontaneous breathing was good and vital signs were normal, without hypoxia. An NGT could not be successfully advanced. A combined chest and abdominal X-ray film showed that the NGT was located in the upper esophageal pouch (Fig. 2A). Moreover, vertebral abnormalities, mediastinal shift and a large gastric bubble without any distal bowel gas were noted. These findings suggested that the infant exhibited EA/TEF complicated with duodenal obstruction and hypoplasia of the right lung. Emergency surgery was performed on the day after birth, confirming the duodenal obstruction due to an annular pancreas (AP). The intra-operative pathological anatomy showed EA with proximal TEF (Gross type C). An extrapleural operation for repair of EA/TEF, and laparotomy and duodenoduodenostomy for repair of AP (EA repair followed by AP repair) were synchronously performed in 2.5 h without gastrostomy. On day 9, enteral feeding was started, and the infant was able to receive full volume oral feeding by day 14. The NGT was removed on day 8, and oral feeding was initiated on day 9 after the swallow test, which indicated no esophageal or duodenal leakage. Repeated upper gastrointestinal imaging confirmed good patency of the esophageal and duodenal anastomosis (Fig. 2B).

The patient was in good health at the 10-month follow-up. An angiogram revealed only mild narrowing at the site of the anastomosis. During the next 15 months, the narrowing progressed into a recalcitrant stricture in spite of multiple endoscopic dilatation procedures. Due to near-complete esophageal obstruction (<2 mm; Fig. 2C), the parents of the patient refused stent placement. Therefore, the patient was a candidate for segmental resection/anastomosis. A detailed discussion on thoracotomy with attempted segmental resection and anastomosis was conducted with the parents; however, they refused the procedures due to the risk of restenosis. Ultimately, following medical evaluation, the patient received an endoscopy-guided magnetic esophageal compression stricturoplasty.

Ethical approval. The two infants received treatment at the Department of Pediatric Surgery of The Northwest Women's and Children's Hospital (Xi'an, China). All clinical application protocols for the techniques performed were approved by the Ethics Committee of The Northwest Women's and Children's Hospital, and were in accordance with the relevant guidelines and regulations. Written informed consent was obtained from the parents of each infant with regard to use of the novel technique. Retrospective Institutional Review Board approval of The Northwest Women's and Children's Hospital was obtained for the purposes of publication.

Treatment of the refractory stricture

Deployment of magnetic ring device. Magnetic rings prepared from a neodymium-iron-boron (Nd-Fe-B) alloy were obtained from the Northwest Institute for Nonferrous Metal Research and airbrush coated with titanium oxide (5-6-μm thick; Fig. 3A). The rings were developed such that they had an outer diameter of 10 mm and a height of 5 mm, with strength of 0.25 T force, and they were placed with suction power between the esophageal compression stricturoplasty.

Endoscopy-guided magnetic esophageal compression stricturoplasty. Sterilized magnets were placed through the transoral approach and via gastrostomy under fluoroscopic and endoscopic guidance (16,17). One magnet ring (mother ring) with an 8F gastric tube was placed in the proximal esophagus using a transoral approach, which reached the stomach cavity through the stenotic segment under endoscopic guidance (Fig. 3B). Subsequently, the gastric tube was placed through the central hole of the daughter ring by gastrostomy. Next, the daughter ring was fixed onto the tube, which was positioned in the stenotic distal esophageal lumen. Magnets were kept

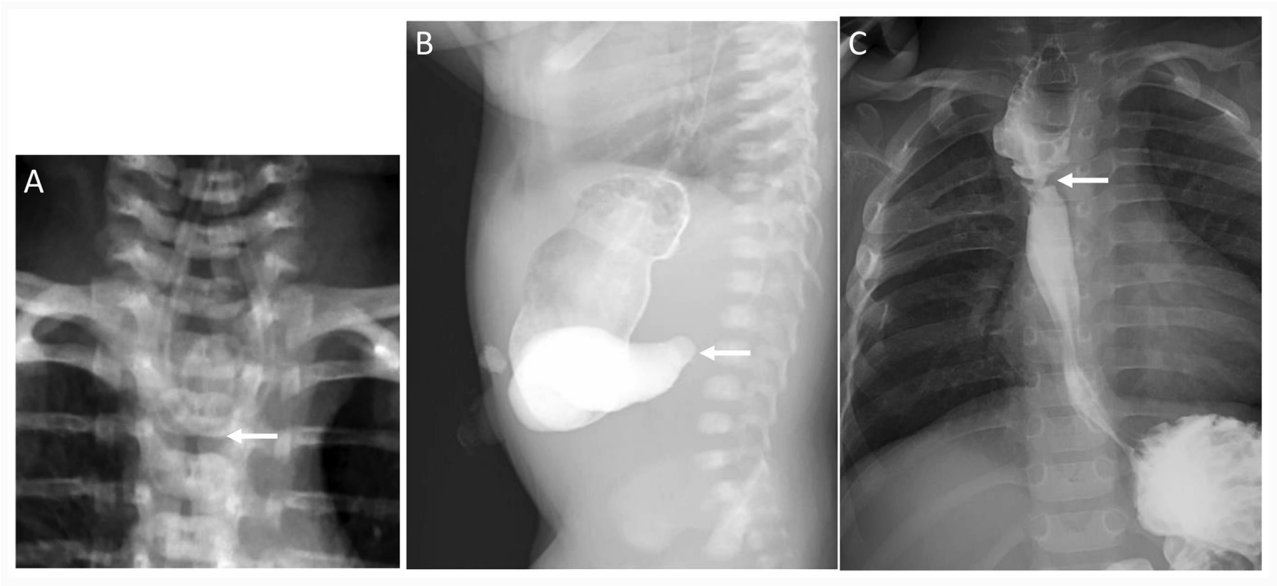

Figure 1. X-ray analysis of patient 1. (A) A positive X-ray confirmed coiling of the nasogastric tube in the upper esophageal pouch (arrow). (B) Upper gastrography confirmed the patency of esophageal anastomosis after the first stage operation and demonstrated a large dilated stomach with duodenal obstruction (arrow). (C) Recurrent anastomotic stricture in the esophagus with <3-mm inner diameter was observed by esophageal radiography at 12 months during the follow-up period (arrow).

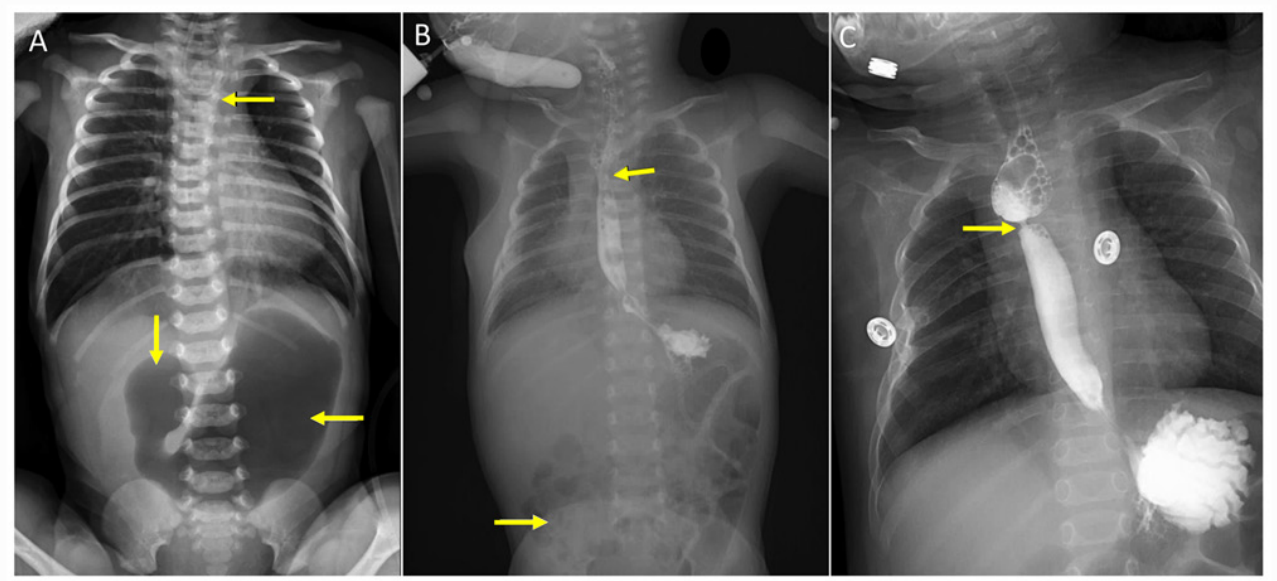

Figure 2. X-ray analysis of patient 2. (A) A preoperative X-ray film showed the combined coiling of the nasogastric tube in the upper esophageal pouch (yellow arrows) and a large gastric bubble with no distal bowel gas (yellow arrows). (B) Upper gastrointestinal imaging confirmed patency of esophageal (yellow arrows) and duodenal anastomosis with distal bowel gas (yellow arrow). (C) Esophageal radiography confirmed esophageal anastomotic stoma stenosis with <2-mm inner diameter at 15 months during the follow-up period (yellow arrow).

in place for 18 days to allow for gradual compression stricturoplasty/anastomosis. Then, under the effect of the magnetic force, the two magnet rings were pulled along the gastric tube to ensure the adequacy of esophageal stricturoplasty. The patient was fed via a gastric tube after the operation.

Outcomes. The placement of magnets was successful in both patients. After the operation, the infants were kept and observed in the Pediatric Intensive Care Unit until the magnets were removed. The magnets achieved full approximation in these two cases, as visualized by a chest radiograph on day 1 (Fig. 4A and C). Attempts were made to move the magnets by gently pulling the tube fixed to the magnets. This process was attempted daily, starting within 1 week post-operation. When the pair of rings could slide up and down, the patients were transferred to the Department of Radiology to confirm the direct apposition of the magnets, and then the magnets were removed under fluoroscopic guidance (Fig. 4B and D). On removal, an upper gastroenterography demonstrated a substantially increased luminal diameter. Furthermore, 8.5- and 9.1-mm anastomotic stomas were formed without esophageal perforation in patients 1 and 2, respectively, with

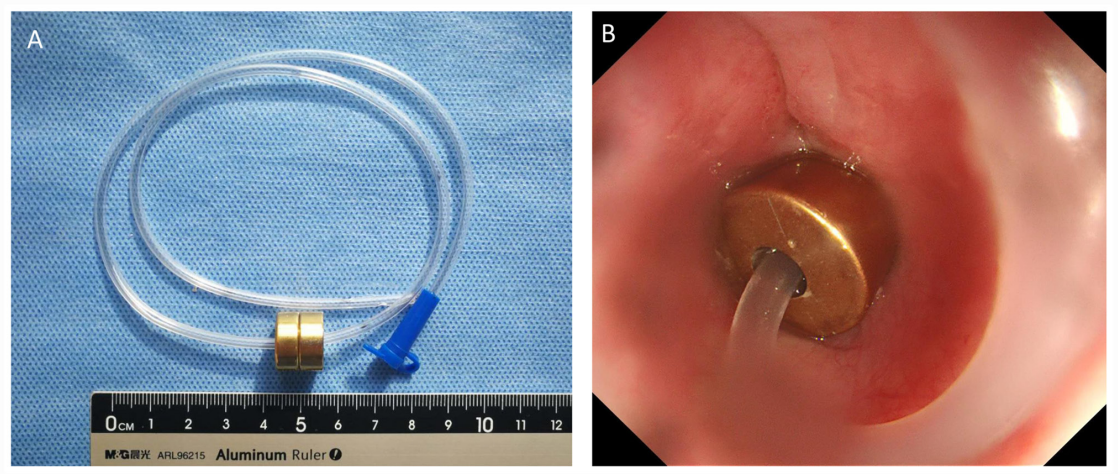

Figure 3. Magnetic compression stricturoplasty process. (A) Magnets were prepared for the operation. (B) Magnets were placed using endoscopy and fluoroscopy.

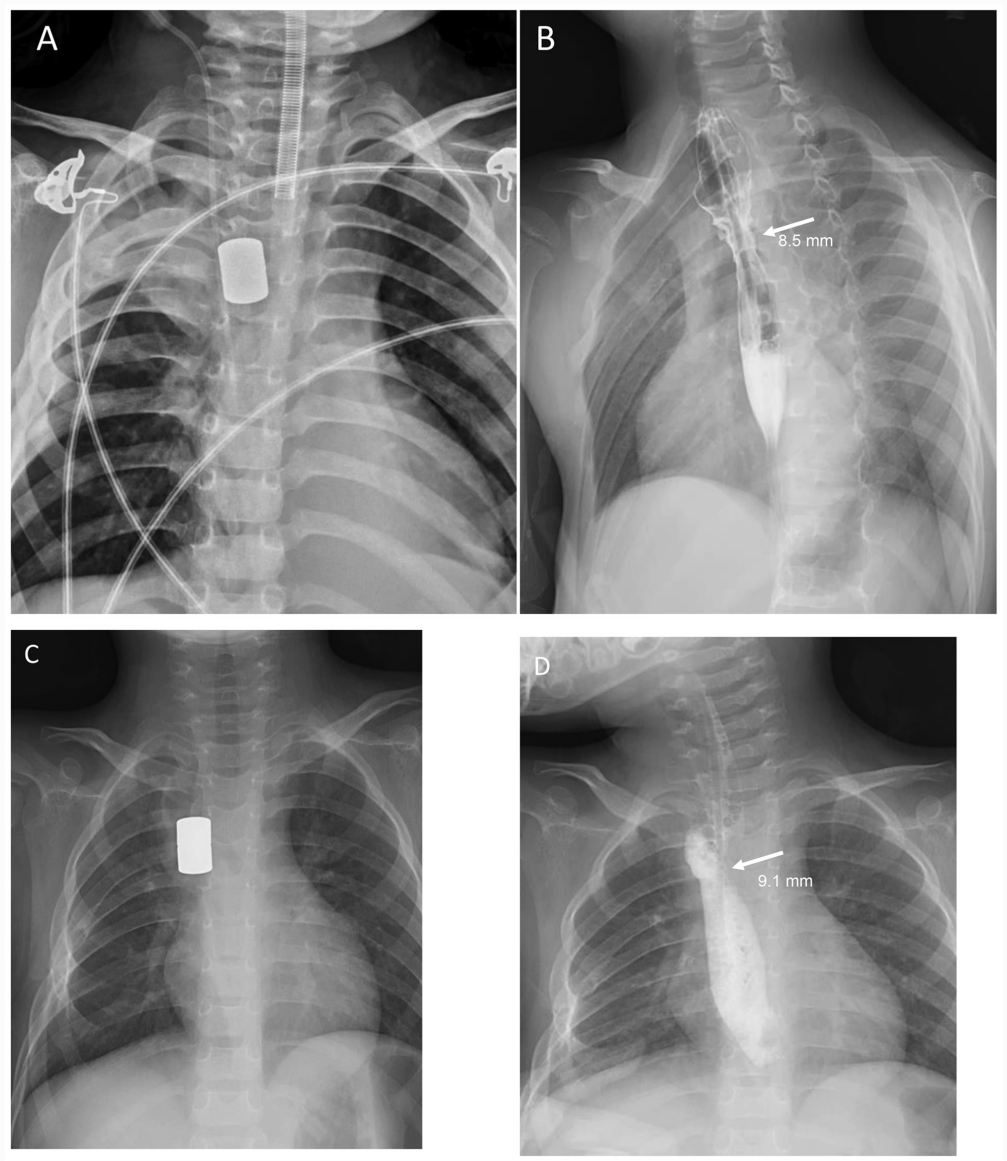

Figure 4. Chest radiography for the two patients. (A) Radiographic approximation was performed on day 1 for patient 1. (B) Magnets were removed and radiographic examination of the patient after magnetic compression stricturoplasty showed a patent esophageal lumen with an inner diameter of 8.5 mm at the anastomotic site on day 14 (arrow). (C) Radiographic approximation was performed on day 1 for patient 2. (D) Magnets were removed on day 18 and a widened lumen was maintained (an anastomotic stoma with an inner diameter of 9.1 mm; arrow).

no other early complications. There was no requirement for balloon dilatation and temporary stent placement after magnetic compression stricturoplasty. At 10 and 15 months after magnetic compression stricturoplasty, no further signs of esophageal stricture were observed. Routine examination showed very good growth and development without any further symptoms of dysphagia or reflux. The follow-up esophagram showed no evidence of residual stricture.

Discussion

Duodenal obstruction is associated with EA/TEF, with a prevalence of ~6% (18). This combined abnormality is associated with high morbidity and mortality rates (1,19). Varying treatment protocols for combined EA/TEF and duodenal obstruction have been reported in the published literature (18,20,21), with high overall prevalence of gastrointestinal morbidity. This may have been due to the concurrent and synergistic effects of various functional and anatomical upper gastrointestinal defects characterizing the two malformations (22).

Due to the rarity of combined EA/TEF and duodenal obstruction, there is a lack of consensus concerning the optimal treatment strategy for these combined abnormalities. Based on the first case reported in the present study, an early combined chest and abdominal X-ray investigation is helpful in avoiding a missed diagnosis of duodenal obstruction in patients with EA/TEF.

In cases similar to the second case reported in the present study, initial thoracotomy with ligation of the fistula and esophageal repair via extrapleural separation should be performed. This may prevent the risk of aspiration and permit the continuation of the laparotomy under stable ventilatory status.

Based on the cases reported in the present study, we propose that in an infant with stable vital signs, a primary simultaneous EA and duodenal obstruction repair should be attempted (4). Surgical treatment of EA/TEF is still maturing, and thoracoscopic ligation has been increasingly performed over the past years (20). The greatest efforts have been made to perform operative treatments of several malformations with the combination of thoracoscope and laparoscope.

A recalcitrant esophageal anastomotic stricture following EA repair in infants presents a surgical challenge, and supportive therapies in combination with balloon or bougie dilatation have been used. However, in a few cases, severe anastomotic stricture may be recalcitrant in spite of several dilatation procedures. The techniques used to treat recalcitrant stricture include intralesional steroid injection and local application of mitomycin C over the stricture site to prevent stricture reformation after dilatation via inhibition of fibroblast proliferation and collagen synthesis (23-25). Additionally, there are reports of using self-expanding stents to prevent stricture recurrence after sufficient dilatation and esophageal stenting (11,12,26). Unfortunately, these traditional methods are not able to effectively remove scar tissues. If bougie or balloon dilatation and these supportive treatments fail, therapeutic options are limited to operative resection of the strictured segment along with reanastomosis or esophageal replacement (13). In past decades, it has been reported that magnetic compression anastomosis (MCA) can be used for benign biliary strictures (27-29), magnetic connectors for coronary surgery (30), functional undiversion of ileostomy (31) and rectal anastomosis (28) in pediatric patients. In pediatric surgery, Russell *et al* (32) determined the effectiveness of MCA in animal models. Zaritzky *et al* (33) first proposed the application of magnets in treating patients with long-gap EA. Takamizawa *et al* (16) reported the application of magnetic compression revision anastomosis in a 31-month-old child who had an anastomotic stenosis following esophagoesophagostomy for long-gap EA without any fistula. Zaritzky *et al* (15,33) reported successful application of MCA in infants who had EA without airway fistula and a gap no broader than 3 cm. Anastomotic stenosis developed in 8 out of 14 patients; among them, two cases needed stent placement and one case needed surgical reanastomosis. Additionally, 5 out of 14 infants received surgical correction of EA, but developed severe recurrent postoperative esophageal stenosis, with no response to dilatation. The patients subsequently underwent MCA, and esophageal reanastomosis was attained within a mean duration of 6 days. In the present study, Nd-Fe-B alloy magnetic rings were prepared with airbrush coating using titanium oxide to enhance their ability to resist gastric acid corrosion, in contrast with previous reports (15,16,33).

In the two cases reported in the present study, discussions were conducted with the parents of each patient regarding alternative treatment options over a time period of weeks before trying the magnetic compression stricturoplasty. These infants had failed standard treatment with endoscopic balloon dilatation, while their parents refused alternative treatments, such as topical injection, stent placement or thoracotomy combined with attempted segmental resection and anastomosis, due to the risk of restenosis. It was hypothesized that, for these two patients, the potential risks associated with surgery were highest for magnetic compression stricturoplasty, in comparison with the other surgical alternatives, which would be less invasive than segmental resection or esophageal replacement. In comparison with the other studies, these two patients did not undergo immediate expansion after magnetic anastomosis. Additionally, it was considered that attempting a magnetic compression stricturoplasty would not preclude future attempts at performing segmental resection or esophageal replacement.

A combination of radio-opaque markers, wires, endoscopy and fluoroscopy were used to achieve the correct orientation of the magnets in the present study. Due to the 'on/off' behavior of magnets through a gap, the surgeon needed to take meticulous care in maintaining the magnet polarity and orientation. Another important property of magnetism is related to the association between attractive force and magnet separation. Importantly, the attractive force between two magnets rises exponentially with reduced separation distance. Considering this property of magnetism, magnetic compression would be well suited for patients who have strictures due to the intrinsic resistance induced by scar tissue. Therefore, it is hypothesized that the interposed resistance caused by scar tissue may delay magnetic coupling, which would lead to a slower/longer process, allowing for stretching of the healthy esophageal segments. In clinical practice, it was observed that magnetic coupling occurred faster than expected. The patients did not develop any leakage and perforation, most probably due to the protection provided by the adjoining scar tissue due to previous surgery.

Esophageal continuity was attained after magnetic compression stricturoplasty for recalcitrant esophageal anastomotic stricture after EA with AP repair. No short-term complications were noted. These findings suggested that this technique is feasible for selected patients. However, there are some limitations of the present study. Firstly, the case number was limited, and additional clinical cases and experience are required to ascertain the necessity and optimal duration of magnetic compression stricturoplasty. Moreover, following the magnetic compression stricturoplasty, the two patients attained esophageal continuity. However, restenosis was a continuing problem, and the patients needed several interventions, including balloon dilatation and temporary stent placement. At 10 and 15 months during the follow-up period, the patients showed durable esophageal patency without dysphagia upon esophagram and clinical examination.

The present case report shows that the early combination of chest and abdominal X-ray investigations can be helpful for treating EA, and may avoid delayed diagnosis of DA. A synchronous operation for repairing EA/TEF and performing duodenoduodenostomy in a single surgery without gastrostomy is recommended for treating the combination of EA/TEF and duodenal obstruction. Magnetic compression stricturoplasty successfully established the patency of the esophagus in these two patients with refractory EA stricture. These two cases required multiple additional procedures, but durable esophageal patency with absence of dysphagia was achieved at 15 or 10 months after magnetic compression stricturoplasty. Further in-depth investigation and follow-up will determine the long-term success of this method. In addition, knowledge of primary magnetic principles will allow for the future customization of magnet arrays for the presentation of individual patients.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

SL conducted the operation, collected data, performed data analysis and interpretation, and drafted the manuscript. YL and JZ designed operation procedures and revised the manuscript. RXL assisted in magnetic ring processing. YF and HY contributed to the endoscopic procedures. RGL and AZ provided assistance in the operation and analyzed and interpreted the patient data regarding the hematological disease. JC and YS contributed to the analysis of the test data in the Intensive Care Unit. NJ contributed to the acquisition and analysis of data regarding the localization of magnets in the esophagus during the anesthetic management of the patient. YL, JZ and SL confirm the authenticity of all the raw data. All authors read and approved the final manuscript.

Ethics approval and consent to participate

All clinical application protocols for the techniques performed were approved by the Ethics Committee of the Northwest Women's and Children's Hospital, and were in accordance with the relevant guidelines and regulations. Written informed consent was obtained from the parents of each infant with regard to use of the novel technique.

Patient consent for publication

Patient consent forms were obtained from the parents/guardians of the patients, giving their consent for the images and other clinical information to be reported in the journal. The parents/guardians understand that the names and initials of the patients will not be published and due efforts will be made to conceal their identities.

Competing interests

The authors declare that they have no competing interests.

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