Staged repair of esophageal atresia: Pouch approximation and catheter-based magnetic anastomosis

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ABSTRACT

Esophageal atresia (EA) without tracheoesophageal fistula is characterized typically by a long gap between the discontinuous upper and lower esophageal pouches. In these challenging patients, initial esophagostomy is often not possible due to excessive tension produced when approximating the two pouches. This tension poses significant risk for pouch retraction, anastomotic leak, mediastinitis, and subsequent esophageal stricture. Consequently, multiple strategies exist to induce pouch lengthening and to achieve full continuity of native esophageal tissue, which if not accomplished, requires interpositions of stomach, small bowel, or colon to establish deglutition. One strategy involves a two-staged approach: first, after a period of growth into early infancy, the two esophageal pouches are suture-approximated without anastomosis, and second, repeat surgery is performed to establish luminal continuity once tension between the pouches has resolved. Esophageal growth and continuity are thereby achieved. Here, we describe an innovative, staged treatment approach for long-gap EA in two premature babies who initially underwent suture-approximation of the esophageal pouches at 3 months of age, followed by catheter-based magnetic compression anastomosis 10 weeks later. In both babies, the magnetic procedure allowed preservation of the gained full esophageal length, established successful esophageal canalization, created a leak-free anastomosis, and prevented the need for additional thoracotomy to accomplish these goals.

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Esophageal atresia (EA) without tracheoesophageal fistula is a rare congenital anomaly [1–3]. The principal goal in the correction of EA is to achieve primary anastomosis of the native esophageal blind ends, or pouches, when possible. The principal challenge to establish esophagostomy is the length of the gap between these pouches. Constructing an anastomosis between non-approximated pouches imparts excessive tension to the repair, which generates a risk of anastomotic leak [4,5] and subsequent mediastinitis, necessitating additional procedures [4,5]. Multiple surgical techniques exist to achieve primary esophageal continuity without interposition of various conduits; however, there is no consensus for the ideal surgical repair in patients who have long-gap EA [5]. Nevertheless, most surgeons believe that inducing and preserving a full-length esophagus will achieve superior results compared to the often technically challenging gastrointestinal interpositions [5,6].

For infants having long-gap EA, in whom it is difficult to achieve an initial, reliable, and safe primary anastomosis, a staged repair is commonly planned. Staged esophageal restoration can take many forms and generally involves an early feeding gastrostomy, followed by either suture-approximation of the two esophageal pouches without anastomosis or suture placement in each of the pouches, which are then transferred through the chest wall for gradual tension-induced lengthening of the esophagus (e.g., the Foker technique) [7]. Historically, both staged approaches have mandated repeat thoracotomy or thoracoscopy to establish esophageal luminal continuity. Staged repairs may also utilize Bakes dilators for repeat noninvasive stretching and lengthening maneuvers that eventually allow approximation of the two
esophageal pouches for delayed primary esophagoesophagostomy. If a long gap persists after induction of esophageal growth, suture-approximation of the two blind esophageal ends (without anastomosis) may be performed, with a surgical anastomosis planned at a later date.

Interestingly, magnetic compression anastomosis has been applied as a primary, minimally invasive alternative to the conventional surgical repair of EA [8,9]. Upon placement of specially designed, latex-free, catheter-based magnets (Cook Medical, Winston-Salem, NC; device not currently marketed) in both blind esophageal pouches, the forces between the two magnets gradually induce pouch growth without interruption of the esophageal blood supply. Minimal tension is exerted during this process. These catheter-based magnets have a maximum outer diameter of 20 French (to allow easy passage through the gastrostomy tract) and taper to a 10 French coupling surface to allow gradient compression anastomosis. Additionally, the upper pouch catheter contains a feeding gastrostomy lumen. Once the magnets connect (i.e., couple), the intervening tissue is compressed, becomes ischemic, and then sloughs centrally, while the outer anastomotic rim heals. This minimally invasive intervention may overcome the traditional obstacles of esophageal anastomosis in babies with EA who cannot reliably undergo initial thoracotomy or thoracoscopy for a primary repair. Through the U.S. Food and Drug Administration Expanded Access Program’s provision for emergency use of an unapproved medical device, we report our preliminary evaluation of magnetic compression anastomosis in two babies who had previously undergone suture-approximation of the two blind esophageal pouches but who still required anastomosis for esophageal continuity.

1. Case reports

1.1. Baby 1

An 890 g triplet girl was born at 27 weeks gestation having a Type A, long-gap EA and also having a rectovestibular fistula, a right aortic arch, and mild tethering of the spinal cord. Three days after birth, the baby underwent insertion of a feeding gastrostomy and a divided sigmoid colostomy. Her siblings (one monozygotic sister and one dizygotic brother) were healthy and had no congenital malformations, consistent with most previous reports that non-genetic risk factors play a role in the development of EA, even in monozygotic multiple gestation siblings [10].

Bakes dilators were used to perform stretching of the esophageal and gastric pouches under fluoroscopic guidance beginning at two months of age (Fig. 1A). The initial gap at this time measured 1.3 vertebral bodies in the anterior to posterior projection. By three and a half months of age, fluoroscopic images suggested that the pouches could be approximated using Bakes dilators within 5 mm and now having an estimated gap distance of 0.3 vertebral bodies (Fig. 1B). The baby then underwent a left retropleural thoracotomy (due to the right aortic arch) for an anticipated primary esophagoesophagostomy. Therefore, the esophageal pouches were approximated with three sutures (one full-thickness stitch connecting the opposing pouch centers and one full-thickness stitch connecting each lateral pouch edge) to promote continued esophageal lengthening and reduction in tension over time.

Two months later, repeat fluoroscopic imaging (calibrated with Bakes dilators) demonstrated that the pouches remained approximated (Fig. 1C). To determine the best means to canalize the two esophageal lumens, the pouches were evaluated endoscopically per os and per gastrostomy. Light from both endoscopes could be visualized from the opposite pouch, but a dilator could not pass easily or safely through the central sutured area due to the thickness of the esophageal walls and scar tissue at this location. To facilitate central canalization and to spare the baby repeat thoracotomy, magnetic compression anastomosis was planned.

After parental consent and IRB approval for emergency use of an unapproved device, the baby was placed under general anesthesia, and repeat endoscopy was performed to re-evaluate pouch approximation (given that four weeks had transpired to plan, design, and acquire approval to perform magnetic compression anastomosis). Briefly, Kumpe catheters were positioned into both esophageal pouches using fluoroscopic guidance, and 0.018-inch wire guides were introduced through both catheters. After removal of the lower Kumpe catheter, the gastric magnetic catheter was placed over the wire guide, and the magnet was positioned at the cephalad portion of the lower esophageal pouch. The remainder of the gastric catheter was pulled back so that the retention balloon...
remained in the stomach. The retention balloon was inflated with 5 mL of saline, and the external bolster was lowered to contact the abdominal skin. Next, using fluoroscopic guidance, the upper esophageal magnetic catheter was inserted over the wire guide and advanced to the tip of the pouch. The magnets appeared to attract immediately and strongly (Fig. 2A and B). A post-procedure X-ray demonstrated a 0.7 mm gap between the magnets. The entire procedure, including endoscopic reevaluation of the pouches, required only 42 min; however, catheter placement alone was accomplished in 14 min. Notably, the esophageal catheter provides a means to suction saliva from the upper pouch, and the gastric catheter also serves as access to provide enteral feedings while the magnets remain in situ. Repeat radiographs demonstrated that on post-procedure day 1, the magnets were 0.4 mm apart and by post-procedure day 3, the magnets were coupled. The magnetic catheters were left in place for an additional three days to allow for compression anastomosis. Before magnetic catheter removal, contrast was injected into the upper esophageal catheter; no anastomotic leak was observed, and luminal continuity was confirmed (Fig. 2C). To facilitate removal, the inner gastric guiding catheter was cut at its proximal end, the retention balloon was deflated, and the gastric catheter was removed from the gastrostomy site. A button gastrostomy tube was then re-inserted. The upper esophageal catheter was removed (along with the coupled magnets) and replaced with an orogastric tube. Between the coupled magnets was a small amount of necrotic compressed esophageal tissue containing a silk suture from the original approximation procedure. Catheter removal was accomplished in 20 min. Upon catheter removal, the baby experienced immediate resolution of hypersalivation and swallowed all oral secretions without difficulty. Five days after catheter removal, an esophogram confirmed no anastomotic leak. Repeat esophagogram, obtained 6 weeks after magnetic catheter removal, demonstrated an anticipated tight but patent narrowing at the anastomosis (i.e., 10 Fr coupling surface of magnets), which was dilated with a balloon to full luminal caliber (Fig. 3A and B). As a routine means to ensure luminal integrity, esophageal dilators were passed weekly for three consecutive weeks. At the time of publication (nearly 8 months after magnet removal), the patient was swallowing salivary secretions well and progressing with oral feedings.

### 1.2. Baby 2

A 36-week gestation female singleton, who had prenatal diagnoses of an open neural tube defect (ONTD), hydrocephaly, and recurrent polyhydramnios, was delivered via Caesarian section and weighed 2940 g. ONTD and hydrocephaly were confirmed postnatally, and additional diagnoses of EA (without tracheoesophageal fistula), neurogenic bladder, and talipes equinovarus were made. ONTD closure and ventriculoperitoneal (VP) shunt placement were performed in the first week of life, and at 8 days of age, a Stamm feeding gastrostomy tube was inserted. A regimen of esophageal stretching with Bakes dilators was implemented similar to that of the first infant (Fig. 1D and E). Again using Bakes dilators to stretch the two pouches manually, the initially estimated gap distance was 1.2 vertebral bodies that gradually reduced to less than 0.3 vertebral bodies over a 2-month span. Due to the VP shunt coursing along the right anterolateral chest wall, a transpleural thoracoscopic approach was selected for this patient to approximate the two esophageal pouches using three interrupted polyglactin sutures. Absorbable suture was used for Baby 2 since the silk suture used for Baby 1 persisted and was found between the magnets upon catheter removal. Notably, significant tension persisted between the two esophageal pouches and obviated any safe attempt to perform primary esophagoesophagegastomy. Given the resulting tension, this suture-approximation procedure was complicated by development of a hydropneumothorax on postoperative day 7, for which a chest washout and re-exploration of the sutured esophageal ends was performed to determine the

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**Fig. 2.** Fluoroscopic images taken at time of catheter-based magnet placement for both babies. A) and D) are anteroposterior projections and B) and E) are lateral projections. C) shows Baby 1 on magnet day 6; the baby was taken to the fluoroscopy suite to assess canalization of the esophageal lumens. Free-flow of contrast through the upper catheter is shown (arrows). The magnets were removed, and an orogastric tube was placed under fluoroscopic guidance. F) shows Baby 2 on magnet day 10; the magnets migrated proximally but were still coupled [arrow, note lower position of magnets compared to D) and E)]. The catheters were removed completely at the bedside, and an 8 Fr orogastric tube was placed until dilation could be initiated the next day.
source of the salivary leak. Interestingly, the esophageal pouches remained nicely approximated without separation, gross ischemia, or visible perforation. Further, no leak could be detected with insufflation of the upper pouch. As a result, two additional polyglactin sutures were placed to reinforce the previous suture line, despite the two esophageal ends appearing otherwise healthy. Two months after this re-operation and spontaneous healing of the salivary fistula, pouch proximity was again assessed with Bakes dilators, as for Baby 1 (Fig. 1F).

After parental consent and IRB approval for emergency use of an unapproved device, placement of the magnetic catheters followed the procedural protocol as described for Baby 1 (Fig. 2D and E). The entire procedure, including endoscopic re-evaluation of the pouches, was completed in 19 min; catheter placement alone was accomplished in 13 min. On day 9 after catheter-based magnet placement, the baby was noted to have scant drainage of saliva via her upper pouch suction tube, suggesting an established communication between the esophageal pouches. Chest radiograph on the following day showed that the magnets, while remaining coupled, had moved proximally in the esophagus, suggesting the anastomosis was complete (Fig. 2F). Plans were then made to remove the magnets on the following day in the fluoroscopic suite, however, the baby pulled the oral catheter (with the magnets still coupled) up into her mouth. The upper and lower catheters and coupled magnets were then removed completely at the bedside without adverse incident; a button gastrostomy tube and a 5 Fr nasogastric feeding tube were placed to maintain access until the routine dilation schedule could be initiated. The baby underwent balloon dilation the next day for the expected anastomotic stenosis (Fig. 3C and D) and had placement of an 8 Fr orogastric tube to facilitate a routine dilating regimen over four consecutive weeks (to achieve proper luminal diameter) as performed for Baby 1. At the time of publication (four months after magnet removal), this baby continues to swallow oral secretions well but has persistent stenosis likely related to the fibrotic healing response of the salivary leak that complicated her original suture-approximation procedure.

2. Discussion

This two-stage approach for correcting long-gap EA described herein can overcome the key challenges encountered during traditional surgical repairs. The first stage suture-approximation procedure encourages lengthening of the native esophageal pouches, while the second stage catheter-based magnetic procedure conserves the entire newly gained esophageal length and establishes luminal continuity through magnetic compression anastomosis. The minimally invasive magnetic procedure further permits a precise and central canalization of the esophageal lumens that is subject to random and imprecise access using other existing methodologies after suture-approximation only. Moreover, in these two babies, magnetic compression created a leak-free anastomosis within 6 and 10 days, respectively, and prevented the need for repeat thoracotomy in the second stage to establish luminal continuity. Placement and removal of the catheter-based magnets were rapid and easily tolerated procedures that did not require additional anesthesia; however, additional sedation or anesthesia can be administered at the physician’s discretion. Because this application of magnetic compression to previously sutured esophageal pouches was a novel endeavor, we believed it was ideal to assess the esophageal pouch proximity, tissue health, and integrity first using flexible endoscopy. Because no abnormalities were noted during endoscopic evaluation of the esophagus of either infant, we believe endoscopy is not a required preliminary procedure; however, such an evaluation may be considered based on physician judgment and/or other anatomic considerations.

The catheter-based magnetic device was initially described as a first-line and single-stage approach to treat patients who had Type A EA or other types of EA in whom any associated

Fig. 3. Fluoroscopic images taken after magnetic compression anastomosis for both babies. A) and B) show an anastomotic stricture before and after 12 mm balloon dilation, respectively, in Baby 1 six weeks after magnetic compression anastomosis. After magnetic catheter removal, Baby 2 self-removed her nasogastric tube the day after its placement (11 days after magnetic compression anastomosis). C) and D) show her anastomotic stricture before and after 10 mm balloon dilation, respectively.
The most recent babies were the catheter-based magnets as described previously, but these two undergo magnetic compression anastomosis for EA applying the same coupling surface), a dilating regimen should be anticipated and the diameter of the magnetic device is small (20 Fr outer, 10 Fr at the balloon dilation or further intervention [8,9]. Notably, because the residual esophageal stenosis or developed stricture that required feeding gastrostomy channel (much like a gastrojejunostomy tube) lumen to aspirate oral secretions, and the lower catheter contains a compression anastomosis, the upper catheter contains a suction importance, as the magnets remain in place actively forming the and precisely the approximated esophageal pouches. Of additional device provided a reasonable solution to align and canalize safely, thereby may misalign the ends. To optimize accuracy of the second stage in the posterior mediastinum, and back into the lower lumen, and since instrumentation may pass out of the upper esophageal lumen maneuvers to bridge the blind pouch ends, however, are imprecise, into the posterior mediastinum, and back into the lower lumen, and thereby may misalign the ends. To optimize accuracy of the second stage in this approach, placement of the catheter-based magnetic device provided a reasonable solution to align and canalize safely and precisely the approximated esophageal pouches. Of additional importance, as the magnets remain in place actively forming the compression anastomosis, the upper catheter contains a suction lumen to aspirate oral secretions, and the lower catheter contains a feeding gastrostomy channel (much like a gastrojejunostomy tube) to provide full enteral nutrition. These cases represent the tenth and eleventh patients to undergo magnetic compression anastomosis for EA applying the same catheter-based magnets as described previously, but these two most recent babies were the first to be treated in the United States. In all previous EA patients, magnetic compression anastomosis was successfully achieved; however, 8 of the 9 patients had an expected residual esophageal stenosis or developed stricture that required balloon dilation or further intervention [8,9]. Notably, because the diameter of the magnetic device is small (20 Fr outer, 10 Fr at the coupling surface), a dilating regimen should be anticipated and planned to achieve the final age-appropriate caliber of the native esophagus. Not surprisingly, the two new patients described herein also experienced narrow anastomoses upon magnet removal and underwent subsequent anastomotic dilation regimens. The first baby reported above having this two-stage approach has gained nearly complete luminal caliber of her original 10-Fr sized anastomosis, now approximating the size of her remaining esophagus, and is progressing with her oral intake appropriate for age. The second baby thus far has residual esophageal stenosis that has been more challenging to manage, likely stemming from the surrounding fibrotic response to the salivary leak that complicated her initial suture-approximation procedure. Although the magnets were able readily to canalize the two lumens in her case, in the setting of scar tissue that developed from the prior salivary leak, all layers of the two esophageal ends were likely less mobile to come together completely, which potentially has resulted in the persistent stenosis, highlighting an important limitation of this two-stage approach (i.e., a salivary leak after suture approximation may predispose to persistent scar tissue that limits mobility and therefore integrity of the subsequent anastomosis). Clearly, meticulous efforts should be made to avoid salivary leak at the time of the initial suture approximation procedure. Nevertheless, while the incidence of residual anastomotic stenosis is high and expected after catheter-based magnetic treatment given the small coupling surface (i.e., 10 Fr), most cases are easily treated by dilation and/or stent placement [12]. Moreover, even after open surgical repair, EA patients are predisposed to gastroesophageal reflux and recurrent esophageal stricture [1,13]. Therefore, anastomotic stenosis or late stricture cannot be solely attributed to the small size of the magnet coupling surface. Finally, the nature, motility, and function of the native esophagus in EA infants is not entirely normal regardless of surgical methods, and we cannot yet comment on the long-term peristaltic and orogastric functions after magnetic compression anastomosis. Open surgical correction is always an option for anastomotic stenosis or stricture if primary minimally invasive interventions are not successful.

While conventional EA treatment outcomes vary by center, by operative technique [14], and by operator experience, the availability of a standardized catheter-based magnetic system may provide more consistent clinical results either as a single- or as a two-staged minimally invasive approach to induce growth and maintain the native esophagus without the need for gastrointestinal interposition. However, this possibility should be demonstrated in a long-term observational study.

3. Conclusion

Two infants underwent successful, catheter-based, magnetic compression anastomosis of previously suture-approximated esophageal pouches as part of a staged repair for long-gap EA. The staged procedure successfully preserved all native esophageal tissue, provided successful and precise esophageal canalization, achieved a leak-free anastomosis, and eliminated the need for repeat thoracotomy to achieve luminal continuity. Given the small size of the current magnet coupling surface (i.e., 10 Fr), a scheduled dilating regimen should be planned within four to six weeks after the compression anastomosis is completed.

Informed consent

Written informed consent was obtained from the legal guardians of these two babies to perform the esophageal magnetic compression anastomosis and to report its results. A copy of the written consent is available for review by the Editor-in-Chief of this journal upon request.

Conflict of interest statement

KJ is a full-time employee of MED Institute, Inc., a Cook Group Company. MZ is an advisor for Cook Medical and shares the patent and royalty payments pending catheter-based magnetic device commercialization.
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Cook Medical provided the catheter-based magnetic devices. The decision to publish this case report was left entirely to the discretion of the participating physicians.

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