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Case Series

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Experience with endoluminal vacuum therapy in the management of esophageal anastomotic leakage in newborns with esophageal atresia: A case series

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KEYWORDS

Endoluminal vacuum therapy, Esophagoplasty dehiscence, Esophageal atresia

ABSTRACT

Background: The management of dehiscence of esophageal anastomosis is challenging and requires a multidisciplinary approach. Endoluminal vacuum therapy (EVT) has shown promising results.

Case Presentation: Herein we present the data of two cases with esophageal atresia who developed esophageal anastomotic leakage, during 2021-2022, and who underwent EVT. The first case had 60% dehiscence of the anastomosis, anastomotic leak repair was performed followed by esophageal EVT placement. In the second case, the dehiscence was limited to 10%, and only esophageal EVT was provided. Within four and six days of EVT, upper gastrointestinal endoscopy and fluoroscopy were performed which showed tissue proliferation in situ and the absence of contrast leakage. Both patients were able to resume oral tolerance on the 4th day.

Conclusion: Esophageal EVT has shown promising results in the management of esophageal anastomotic leakage in newborns with esophageal atresia.

INTRODUCTION

Esophageal atresia (EA) repairs can develop severe complications such as esophageal dysmotility (78%), gastroesophageal reflux disease (43%), dysphagia (44%), anastomotic leak (19%), anastomotic stricture (26%), recurrent fistula formation (7%), and esophagitis (47%). [1] Initial management of anastomotic leak includes holding oral feeding, broad-spectrum antibiotics, esophageal decompression with a naos-esophageal tube (low-pressure suction), and external drainage with a chest tube. If these measures fail, the next step is surgical correction. [2]

Vacuum therapy is widely used to treat skin and muscle defects. [3] It decreases the secretions and wound edema, improves microcirculation, induces granulation, and decreases the wound size. [4] Since the 1990s, its endoscopic application has increased in perforations and fistulas of the esophagus. [5] EVT is adopted for esophageal perforations in 2008 and the first pediatric experience of EVT for closure esophageal perforations was in 2018. [2] It is used by placing polyurethane sponges at the defect site and connecting them to an EVT device to achieve the described effects. [6] It also prevents leakage of secretions into the mediastinum.

Manfredi et al. [2] reported that the mean hospital stay was 5.5 days. The success rate, based on the closure of the EVT esophageal perforation, is 88%. [6] Reports on neonates are scarce, so the importance of this work is to support this approach in this particular patient population.

A review of 5 cases with EA type III was carried out in the period 2021 - 2022, in a Pediatric Hospital in Guayaquil - Ecuador; 2 cases developed esophageal anastomotic dehiscence, and both were managed with esophageal EVT for the first time in the hospital. Data were obtained through chart review and surgical records; the respective consent was taken for their report.

Technique

Upper gastrointestinal flexible endoscopy (UGE) is performed under general anesthesia and the distance from the dental arch to the dehiscence is measured. A polyurethane sponge is shaped according to the defect size and attached to a Nelaton catheter 6 or 8 FR with

a braided 2.0 suture and introduced under direct vision with the laryngoscope; once positioned according to the estimation, the flexible endoscopy confirms its position. This device is connected to a continuous negative pressure system. The pressure ranges between 75 and 150 mmHg. A pressure of 75 mmHg is considered low intensity, 125 mmHg of medium intensity, and 150 mmHg as high intensity in pediatric patients [7]; in neonates, we apply 75-100 mmHg, and chest drainage is maintained (Fig. 1).

Post-operative care takes place in the neonatal intensive care unit (NICU), where the patient is kept intubated. Decreasing chest tube output is correlated with the degree of dehiscence closure.

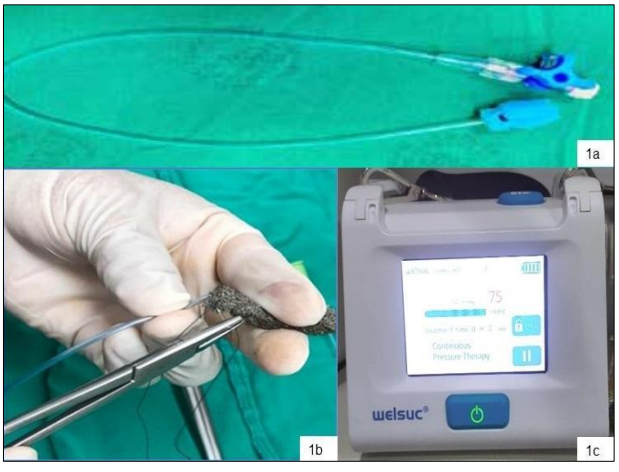

Figure 1: 1a) NGT. 1b) sponge attached to NGT. 1c) NWPT device

An average of between four to six days was considered for the esophageal review, UGE is performed and it begins by instilling 10-20 ml of 0.9% NaCl to avoid injuries since the adhesion between the sponge and the granulation tissue can be firm; If no leaks are observed, fluoroscopy is performed with a water-soluble contrast medium that corroborates the absence of leaks at the anastomosis site. The patient is transferred to the neonatal intensive care unit for post-surgical management. The patients are allowed orally four days after the withdrawal of the EVT. The patients are called for follow-up after 15 and 30 days post-discharge and then every month

RESULTS

Case 1: A term male neonate born through the cesarean section was admitted with a diagnosis of EA (type C). Echocardiography showed Tetralogy of Fallot. Rigid bronchoscopy visualized the fistula located at two tracheal rings above the carina. A right thoracotomy with primary esophageal anastomosis was performed.

On the 4th day of surgery, a right-sided tube thoracostomy was done for pneumothorax. The chest tube was placed under negative pressure. On the 6th postoperative day, saliva was observed in the thoracostomy tube along with remarkable respiratory distress. A decision for thoracoscopy was made, which revealed a dehiscence of esophageal anastomosis of approximately 60% on the posterior side, which was repaired with extracorporeal knots, and endoluminal vacuum therapy was applied with negative pressure set at 75 mmHg. A chest tube was also placed.

After six days, no drainage was seen through the chest tube. The UGE findings were: in-situ granulation tissue and no evidence of leakage (confirmed by fluoroscopy). A trans-anastomotic orogastric tube (OG) was placed and the chest tube was removed. The oral trial was given on the 4th day and the patient was discharged on the 6th day of gastroscopy. The patient is doing fine on 1 monthly follow-up.

Case 2: A term female neonate, born via cesarean section was admitted with type C esophageal atresia. Rigid bronchoscopy visualized fistula at C1. Right thoracoscopy was performed with primary closure (tracheoesophageal fistula) and esophagoplasty. A chest tube was placed and the patient was transferred to the NICU. On the 3rd postoperative day, pneumothorax developed for which tube thoracostomy was done with negative pressure of -5 cmH2O.

On the 8th day, breast milk was started through the OG tube. On the 9th day, dressings were stained and a chest X-ray showed bilateral consolidations. Subsequently, UGE was performed and dehiscence of the anastomosis was confirmed (10%). The endoluminal vacuum therapy was placed, using a 6Fr Nelaton catheter. Negative pressure was set at 75 mmHg. A chest tube was kept in place. The patient was transferred to NICU, still intubated.

UGE findings on the 4th day were: fibrin present in the anastomosis area; esophagogram did not show any leakage. The esophageal EVT device was then removed. In four days an oral trial was started. On follow-up, the patient is doing fine.

DISCUSSION

Surgical repair is the best next step in the traditional treatment of major esophageal leaks in newborns operated for esophageal atresia. EVT is one of the recent advances in the management of esophageal anastomotic leaks in neonates with esophageal atresia. In this case series, we described our experience of the use of EVT in two neonates with anastomotic leakage. In one case the leakage was major (60%) whereas in another it was minor (10%).

The use of customized EVT devices guides the sponge to the correct site under fluoroscopy. The intensity of negative pressure (between 75 and 150 mmHg) was chosen based on provider experience and pre-existing data on VTE in children. [1]

Table 1: Summary of cases presented.

Data	Case 1	Case 2
Sex	Male	Female
Gestation Weeks	38GW/AGA	38GW/ AG
Birth weight	2,8 kg	2,3 kg
Prenatal diagnostic	No	No
Diagnostic	EA type C + TF	EA type C
Surgery	Rigid bronchoscopy (fistula to 2 tracheal rings on carina) Right thoracotomy + chest drainage (tension esophagoplasty)	Diagnostic bronchoscopy (fistula to 3 tracheal rings on carina) + right thoracoscopy + closure of T-E fistula + esophagoplasty + thoracic drainage
Post-surgical pneumothorax	4th day	3rd day
2nd surgery	Video thoracoscopy + anastomosis with extracorporeal knots + EVT + chest drainage (esophageal dehiscence 60%, poor quality esophagus, use Nelaton catheter 8Fr OG a - 75 mmHg	UGE + chest drainage (dehiscence 10%) + EVT-Nelaton catheter 6Fr OG a - 75 mmHg)
Duration of therapy	6 days	4 days
% Sealed	100%	100%
Started orally	4th day after EVT removal	4th day after EVT removal
Follow up	Monthly	Monthly

GW: gestation weeks; AGA: adequate for gestational age; kg: kilograms; EA: esophageal atresia; TF: tetralogy of Fallot; T-E: tracheoesophageal; EVT: endoluminal vacuum therapy; OG: orogastric; UGE: upper gastrointestinal endoscopy.

Table 2. Characteristics of patients with esophageal leak treated with EVT

Data	Total patients	Ages (days)	Therapy duration (days)	Sponges changed	Perforation closure	Post-surgical
Our Data	2	6-8	4-6	1	2/2 (100%)	-
Manfredi et al. [1]	17	210-1140	8 (6-13)	2(1-3)	15/17 (88%)	7/8 (88%)
Kaczmarek et al. [7]	4	0-17	22(7-39)	6(1-12)	4/4 (100%)	-
Ritz et al. [9]	5	210-4080	30(21-45)	5(2-8)	4/5 (80%)	1

In the 1st case, esophagoplasty was performed with extracorporeal knots due to 60% dehiscence. Whereas in the second case as the leakage was minor only EVT was applied without repairing the esophageal leak. To confirm the outcome of this therapy objectively, UGE and fluoroscopy, between days four to six, were performed that showed the absence of leaks, thus helping decide the removal of the esophageal EVT.

During EVT, the patients remained NPO, and parenteral nutrition was provided until the absence of leaks is verified. Fasting is considered in these patients because oral nutrition would contaminate both the perforation site and the sponge, rendering EVT ineffective [7, 8].

Table 2 summarizes the characteristics of the patients (from the literature search) with esophageal leaks treated with TVE, including our cases.

In both cases, all the perforations were healed in comparison with the studies mentioned in Table 2. It should be noted that there are 2 cases compared to the studies mentioned that have a range of patients from 4 to 17 patients; none of our patients needed a post-surgical seal, nor several sponge changes. However, our experience is very limited and studies with large sample sizes and experimental studies are recommended to adopt this technique as a standard therapy for the management of esophageal anastomotic leaks in patients with esophageal atresia.

CONCLUSION

We reported the use of EVT in two patients with esophageal anastomotic leakage. In both patients, the objective of closure of the dehiscence was met. So far there have been no complications and the weight gain has been adequate. We conclude that the use of the esophageal EVT device in neonates with anastomotic dehiscence is technically feasible. This study in neonates is a contribution to an approach to the use of the device. Further prospective studies, with more cases, are needed to fully assess the benefits and complications of this promising approach

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REFERENCES

1. Comella A, Tan Tanny SP, Hutson JM, Omari TI, Teague WJ, Nataraja RM, et al. Esophageal morbidity in patients following repair of esophageal atresia: A systematic review. J Pediatr Surg. 2021; 56:1555-63. Available from: https://doi.org/10.1016/j.jpedsurg.2020.09.010.
2. Manfredi MA, Clark SJ, Staffa SJ, Ngo PD, Smithers CJ, Hamilton TE, et al. Endoscopic esophageal vacuum therapy: a novel therapy for esophageal perforations in pediatric patients. J Pediatr Gastroenterol Nutr. 2018; 67:706–12. Available from: https://doi.org/10.1097/MPG.0000000000002073.
3. Safronov AA. Vacuum therapy of trophic ulcers with simultaneous autoplastic skin transplantation (Article in Russian). Novyi Khirurgicheskii Arkhiv. 1962; 3:3–8.
4. Still S, Mencio M, Ontiveros E, Burdick J, Leeds SG. Primary and rescue endoluminal vacuum therapy in the management of esophageal perforations and leaks. Ann Thorac Cardiovasc Surg. 2018; 24:173-9. Available from: https://doi.org/10.5761/atcs.oa.17-00107.
5. Bludau M, Hölscher AH, Herbold T, Leers JM, Gutschow C, Fuchs H, et al. Management of upper intestinal leaks using an endoscopic vacuum-assisted closure system (E-VAC). Surg Endosc. 2014; 28:896-901. Available from: https://doi.org/10.1007/s00464-013-3244-5.
6. Huang C, Leavitt T, Bayer LR, Orgill DP. Effect of negative pressure wound therapy on wound healing. Curr Probl Surg. 2014; 51:301-31. Available from: https://doi.org/10.1067/j.cpsurg.2014.04.001.
7. Kaczmarek DJ, Heling DJ, Strassburg CP, Katzer D, Düker G, Strohm J, et al. Management of esophageal perforations in infants by endoscopic vacuum therapy: a single center case series. BMC Gastroenterol. 2022; 22:282. Available from: https://doi.org/10.1186/s12876-022-02346-2.
8. Schorsch T, Müller C, Loske G. Endoscopic vacuum therapy of anastomotic leakage and iatrogenic perforation in the esophagus. Surg Endosc. 2013; 27:2040-5. Available from: https://doi.org/10.1007/s00464-012-2707-4.
9. Ritz LA, Hajji MS, Schwerd T, Koletzko S, von Schweinitz D, Lurz E, et al. Esophageal perforation and EVAC in pediatric patients: A case series of four children. Front Pediatr. 2021; 9:727472. Available from: https://doi.org/10.3389/fped.2021.727472.