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OPEN SURGICAL REPAIR OF CONGENITAL ESOPHAGEAL ATRESIA AND TRACHEOESOPHAGEAL FISTULA BY AZYGOS SPRING VERSUS CLASSIC REPAIR

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ABSTRACT

Background: The open EA/TEF repair is a test of surgical skills of a pediatric surgeon. In a baby with EA and a distal TEF, the lungs may be exposed to gastric secretions, air from the trachea can pass down the distal fistula when the baby cries, strains, or receives ventilation. This condition can lead to an acute gastric perforation, which is often lethal. The thoracoscopic approach to repair EA/TEF provides excellent view, allowing the most skillful surgeons to spare the azygos vein by performing the esophageal anastomosis over (on the right side) the azygos vein. **Aim of the study** was to compare a short-term outcome of open surgical repair of esophageal atresia and tracheoesophageal fistula (EA/TEF) by Azygos spring versus classic repair. **Patients and Mehods:** This prospective comparative study was carried out at the Pediatric Surgery Department, at Zagazig University Hospital during the period study year 2017 till the year 2019, 22 neonates diagnosed with EA/TEF were admitted to NICU undergoing azygos spring repair versus 22 NICU neonates diagnosed with EA/TEF undergoing classic repair operations. **Results:** Regarding short-outcome results, leakage, pneumonia, and overall complications were significantly in lower rate with Azygos repair and associated with classic group. **Conclusion:** Open surgical repair of EA/TEF with preservation of the azygous vein and if possible immediate or early extubation, leads to good results if performed by an experienced surgeon with respect to other perioperative factors.

Keywords: Open Surgical Repair, Congenital Esophageal Atresia, Tracheoesophageal Fistula, Azygos Spring

INTRODUCTION

Esophageal atresia (EA) encompasses a spectrum of malformations comprising an interruption of the esophagus, predominately in combination with persistent communication with the trachea (tracheoesophageal fistula, TEF) [(1)].

The open EA/TEF repair is a test of surgical skills of a pediatric surgeon. The postoperative management is also considered a test of standards of neonatal care. The series reported so far have shown the feasibility and comparable (even better) results with the open technique [(2)].

Early and late postoperative morbidities frequently occur after initial repair of EA/TEF despite excellent surgical and neonatal management, and can be associated with impaired outcomes [(3, 4)]. Early complications included anastomotic leaks, recurrent TEF with or without anastomotic stricture, initially missed proximal fistula and refractory anastomotic strictures with or without fistula [(5)].

Anastomotic strictures are one of the most frequent problems after EA/TEF repair and can significantly complicate the further outcome. The majority usually respond well to repeated dilatations with bougies or hydrostatic balloons without need for further intervention [(6)].

AIM OF THE WORK

The aim of this study is was to compare a short-term outcome of open surgical repair of esophageal atresia and tracheoesophageal

fistula (EA/TEF) by Azygos spring versus classic repair.

PATIENTS AND METHODS

This study was carried out at the Pediatric Surgery Department, at Zagazig University Hospital during the period study year 2017 till the year 2019. The study enrolled 22 NICU neonates diagnosed with EA/TEF undergoing azygos spring repair versus 22 NICU neonates diagnosed with EA/TEF undergoing classic repair operations. Written informed consent was obtained from all patients and the study was approved by the research ethical committee of Faculty of Medicine, Zagazig University. The work has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for studies involving human.

Inclusion criteria:

- Newborns with the diagnosis of EA with TEF (cross type C).

Exclusion criteria:

- Newborns with respiratory distress (RD) requiring ventilatory support.
- Newborns with major anomalies.
- Newborns with septic shock.
- Prematurity and/or birth weight of less than 1500 g.
- Birth weight less than 1500 g.
- Long gap necssitiating staged operation

Methods:

Cases were selected with simple random sampling. Data collected retrospectively from hospital and office records. All newborns were subjected to laboratory investigation including CRP before and after operation, echocardiography evaluation, ultrasonography evaluation, regarding age of patients, time of operation, associated anomalies, hospital stay, associated anomalies, associated anomalies, postoperative extubation time, time of oral feeding, leakage if occurred, results of gastrographin study, mortality, pneumonia if occurred provide by CXR.

Surgical techniques:

The primary goal of the initial operation is to divide and close the esophageal fistula. Simple ligation (tying off) of the esophageal fistula is not recommended because of the high risk of recurrence. Once the esophageal fistula is divided an end-to-end esophageal anastomosis should be performed.

In unstable or premature infants, it is often safer to perform a staged repair by performing the esophageal anastomosis at a later date, after the patient has stabilized and grown. If no anastomosis is performed, a gastrostomy tube should be placed at the initial operation for feeding access until the atresia can be repaired. A chest tube is often placed to evacuate any leakage from the esophageal anastomosis.

Through right posterolateral thoracotomy, posterior mediastinum is reached by gently sweeping the parietal pleura off the endothoracic fascia with the help of wet pledgets. Azygos vein is seen crossing from right to left. A nick is made in the endothoracic fascia just below the azygos arch. A blunt right angled forceps is passed beneath the azygos arch and a tunnel is made by stripping off the endothoracic fascia above it.

If the lower esophageal pouch ends in a fistula below the azygos arch, it is identified and the fistula is divided and closed either by interrupted non- absorbable sutures or by transfixation. The lower divided end of esophagus is passed through the tunnel beneath the azygos arch and is anastomosed end to end with the upper esophageal pouch after its adequate mobilization. In case the lower esophageal pouch is ending in a fistula with the trachea at a higher level, it can be hooked above the azygos arch with the help of a gentle downward traction on the azygos arch. The anastomosed neoesophagus lies medial to the azygos vein as in normal subjects.

The surgical procedure was through right thoracotomy in the third or the fourth space, and retropleural dissection to reach the posterior mediastinum by retracting the pleura and the lung medially.

Careful dissection of the azygous vein was performed without ligation. Identification of fistula and transfixation ligature was applied; esophageal continuity was established by end to end anastomosis was done to restore esophageal continuity. Posterior mediastinal drains were inserted close to the repair site. All patients were followed up for leakage, respiratory complications, and/or sepsis.Oral gastrografin imaging was performed if no

leakage was encountered when feeding was started gradually; if leakage occurred, the nothing per oral (NPO) period was increased to 3 or 5 more days, and then leakage was re-evaluated (fig 1S, 2S,3S).

RESULT

The present study enrolled 22 NICU neonates diagnosed with EA/TEF undergoing azygos spring repair versus 22 NICU neonates diagnosed with EA/TEF undergoing classic repair operations. All newborns and infants were subjected to age of patients, time of operation, echocardiography, ultasonography, prenatal conditions before operation, associated anomalies, hospital stay, postoperative extubation time, time of oral feeding and CRP before and after operation.

Table (1), showed Age distribution between groups, where the mean age in azygos group was 5.42±2.9, while in classic repair group it was 5.2±3.1, with no significant difference between groups, Table (2), showed sex distribution among studied group, the azygos group were 13 cases males and 9 cases females, in classic repair group were 12 cases males and 10 cases females with no significant differences between groups regard sex as male/female nearly matched. Table (3) showed CRP distribution between groups where the Mean in azygos group was 44.11±28.0 and it was 36.51±23.84 in classic repair group, and no significant difference according to CRP distribution between groups. Table (4) showed weight distribution between studied group, where the Mean in azygos group was 2.83±0.42and it was 2.80±0.51 in classic repair group with no significant difference according to weight distribution between studied group. Table (5), showed associated anomaly distribution between studied groups, where athe ssociated anomalies was 68.2% in azygos group and 72.7% in classic repair group with no significant difference between groups regard associated congenital anomaly. Table (6), showed exitubation time distribution between studied groups, where it was 22.7% in azygos group and 9.1% in classic repair group with a significant difference for Exitubation time. Table (1S), showed operation time, hospital and ICU stay distribution between studied groups, where the operation time was 54.5±11.83, hspital stay was 9.21±3.23 and ICU stay was 4.51±0.83 in azygos group and the operation time was 57.66±16.35, hspital stay was 11.35±2.87 and ICU stay was 6.88±0.83 with a significant difference in hospital and ICU stay as it was significantly higher in the classic repair group. Table (2S), showed Short outcome results and complication distribution between studied groups, where leakage rate was 18.2% in Azygos spring group and 34.1% in classic group and Pneumonia rate was 13.6% in Azygos spring group and 36.4% in classic group, leakage, pneumonia and overall complications were significantly in lower rate with Azygos repair and associated with classic group, with significant differences between the studied groups. Table (3S), showed **Short outcome results and complication distribution between studied groups, where** the mortality rate was significantly higher in classic group, where the survival rate in Azygous group was 77.3% but in classic group it was 61.4% and the mortality was significantly higher in classic group.

Table 1 Age distribution between groups

		Azygos spring Group (N=22)	Classic repair Group (N=22)	Mann Whitney/t	P
Age	**Mean ±SD**	**5.42±2.9**	**5.2±3.1**	**0.255**	**0.801**
	Median (Range)	**4 (1-13)**	**3 (1-12)**		

Table 2 Sex distribution among studied group

			Group		Total	X^2	P
			Azygos spring repair Group	Classic repair Group			
Sex	Male	N	**13**	**12**	**25**	**0.093**	**0.76**
		%	**59.1%**	**54.5%**	**56.8%**		
	Female	N	**9**	**10**	**19**		
		%	**40.9%**	**45.5%**	**43.2%**		
Total		N	**22**	**22**	**44**		
		%	**100.0%**	**100.0%**	**100.0%**		

Table 3 CRP distribution between groups

		Azygos spring repair Group (N=22)	Classic repair Group (N=22)	Mann Whitney/t	P
CRP	**Mean ±SD**	**44.11±28.0**	**36.51±23.84**	**1.512**	**0.134**
	Median (Range)	**41 (18-100)**	**25 (9-90)**		

Table 4 Weight distribution between studied group

		Azygos spring repair Group (N=22)	Classic repair Group (N=22)	Mann Whitney/t	P
Weight	**Mean ±SD**	**2.83±0.42**	**2.80±0.51**	**0.119**	**0.906**

Table 5 Associated anomaly distribution between studied groups

			Group		Total	X^2	P
			Azygos spring repair Group	Classic repair Group			
Associated anomalies	NO	N	**15**	**16**	**31**	**1.53**	**0.82**
		%	**68.2%**	**72.7%**	**70.5%**		
	ASD	N	**3**	**1**	**4**		
		%	**13.6%**	**4.5%**	**9.1%**		
	FALLOT	N	**2**	**1**	**3**		
		%	**9.1%**	**4.5%**	**6.8%**		
	Undescended testis	N	**1**	**1**	**2**		
		%	**4.5%**	**4.5%**	**4.5%**		
	VSD	N	**1**	**3**	**4**		
		%	**4.5%**	**13.6%**	**9.1%**		
Overall Anomaly	-VE	N	**15**	**16**	**31**	**0.1**	**0.74**
		%	**68.2%**	**72.7%**	**70.5%**		
	+VE	N	**7**	**5**	**12**		
		%	**31.8%**	**27.3%**	**29.5%**		
Total		N	**22**	**22**	**44**		
		%	**100.0%**	**100.0%**	**100.0%**		

Table 6 Exitubation time distribution between studied groups

			Group		X^2	P
			Azygos spring repair Group	Classic repair Group		
Extibation time	Immediate	N	**5**	**2**	**13.88**	**0.007***
		%	**22.7%**	**9.1%**		
	1st	N	**13**	**4**		
		%	**59.0%**	**18.2%**		
	2nd	N	**2**	**4**		
		%	**9.1%**	**18.2%**		
	3rd	N	**1**	**7**		
		%	**4.5%**	**31.8%**		
	No	N	**1**	**5**		
		%	**4.5%**	**22.7%**		
Total		N	**22**	**22**		
		%	**100.0%**	**100.0%**		

DISCUSSION

Esophageal atresia (EA) with or without a tracheoesophageal fistula (TEF) is one of the rare congenital anomalies that occurs in one in 3000 births. Traditionally, these patients present shortly after birth because of an inability to pass a nasogastric tube, respiratory distress, or an inability to tolerate feedings. The condition may be associated with other major congenital anomalies (VACTERL syndrome) or may be an isolated anomaly [(7)].

TEF should be considered a touchstone in pediatric surgery. It demands early diagnosis, resuscitation, and emergency surgical correction. The associated congenital anomalies worsen the outcome [(8)].

Survival rates of greater than 90% are reported; recent studies on infants undergoing thoracoscopic repair have reported mortality rates as low as 3%, with case fatalities often related to associate cardiac or other congenital anomalies [(9)].

The outcome of surgical repair can be evaluated immediately postoperatively, within days or weeks, in terms of mortality. The assessment of late outcomes among surviving cases in terms of morbidity through follow-up, with a focus on gastrointestinal-tract (GIT), respiratory, musculoskeletalrelated complications, and quality of life, requires studies of longer durations. [(10)]

In our study, there were no significant differences between groups regarding age, C-reactive protein and weight; also, there were no significant differences regarding sex (males and females were nearly matched in azygos spring repair and classic repair groups respectively).

Also, there were no significant differences between groups regarding associated congenital anomalies, operation time, hospital stay and ICU stay.

In a study published in 2007, the preservation of the azygos vein during surgery in patients with EA and TEF resulted in a significant reduction in the number of postoperative anastomotic leaks [(11)]. In another study, a reduction in the rate of postoperative pneumonitis was observed with preservation of the azygos vein after surgery for TEF [(12)].

While in our study, leakage rate was 18.2% in Azygos spring group and 34.1% in classic group and Pneumonia rate was 13.6% in Azygos spring group and 36.4% in classic group, leakage, pneumonia and overall complications were significantly in lower rate with Azygos repair and associated with classic group, with significant differences between the studied groups.

Survival rates of greater than 90% are reported; recent studies on infants undergoing thoracoscopic repair have reported mortality rates as low as 3%, with case fatalities often related to be associated with cardiac or other congenital anomalies [(11)].

Rintala et al. [(13)] reported that the mortality rate of infants with EA/TEF has decreased

significantly with improving surgical techniques and perioperative care. Survival rates of greater than 90% are now routinely reported.

While in our stydy, the mortality rate was significantly higher in classic group, where the survival rate in Azygous group was 77.3% but in classic group it was 61.4%.

Causes of mortality in our study was due to the delay before arrival of cases to hospital, the long period taken by the anesthesiologists to prepare cases preoperatively and the limited number of incubators, so cases should wait for a time till there is an empty incubator [(10)].

Elshahat et al. [(10)] evaluated the immediate outcome of open surgical repair of the EA/TEF (Gross C) sparing azygous vein through the retropleural approach. They concluded that opensurgical repair of EA/TEF by the extrapleural approach, with preservation of the azygous vein and if possible immediate or early extubation, leads to good results if performed by an experienced surgeon with respect to other perioperative factors.

El-shahat et al. [(10)] stated that there are multiple factors affect the immediate outcome of EA/TEF repair including neonatal age at the time of operation, weight, C-reactive protein (CRP) level as a predictor for sepsis, operative duration, postoperative intubation period, anastomotic leakage, and hospital stay; all these factors affect the early outcome.

CONCLUSION

- Regarding short outcome result, leakage, pneumonia, significantly in lower rat with azygos spring repair and associated classic group:
- In our study there is no significant difference group regarding age, C-reactive protein, sex (male and female) nearly matched in azygos spring repair and classic repair group.
- There is no significant difference between azygos spring repair and classic repair group according to operation time and congenital anomalies.
- The classic repair was significantly associated with high hospital and ICU stay.
- In our study the extubation give good results if immediately as early as possible was done.
- Regarding to short outcome in our study, ICU and hospital stay was signicantly lower rate azygos spring repair.
- Postoperative pneumonia and leake was lower in azygos spring repair in comparison with classic repair group.

Declaration of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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