Factors affecting the short-term outcome of open surgical repair of esophageal atresia and tracheoesophageal fistula through an extrapleural approach with azygous vein sparing

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Background
Congenital esophageal atresia (EA) and tracheoesophageal fistula (TEF) are common congenital anomalies, affecting one in 2400–4500 neonates. The surgical management of EA/TEF has evolved over the last 60 years and is associated with a progressive improvement in survival.
In this study, we performed open surgical repair of EA with TEF, type (Gross C). In all cases, we proceeded with the azygous sparing retropleural approach and early extubation if circumstances permitted. In all cases, we studied all other factors affecting the results in our institute.
Aim
To evaluate the immediate outcome of open surgical repair of the EA/TEF (Gross C) sparing azygous vein through the retropleural approach.
Patients and methods
In this prospective study, 26 neonates diagnosed with EA/TEF were admitted to NICU and prepared for surgery. All patients were operated on through right thoracotomy with an extrapleural approach and preservation of the azygous vein. Perioperative factors affecting the outcome were evaluated and recorded.
Results
Of the 26 neonates operated on, 15 were males and 11 were females. The mean age of the neonates at operation was 5.5 days. Their mean weight was 2875 g. Twenty-one (80.7%) cases survived and were discharged, whereas five (19.2%) cases died in the early postoperative period because of sepsis, respiratory, and ventilator complications. The mean preoperative C-reactive protein (CRP) level was 112. Leakage as a potential complication occurred in eight out of 26 (30.7%) cases. Immediate extubation was achieved in 12 (46.1%) cases. The operative time ranged from 45 to 100 min and the mean hospital stay was 12 days.
Conclusion
Open surgical 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.

Keywords:
azygos, esophageal atresia, extrapleural, tracheoesophageal fistula

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Introduction

Esophageal atresia (EA) with or without a tracheo-esophageal fistula (TEF) is one of the rare congenital anomalies that occurs in one in 3000 births [1,2].

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 [3].

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 [4].

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 associated cardiac or other congenital anomalies [5,6].

Improvement in survival is multifactorial and largely attributable to the advances in neonatal ICU, anesthetic management, ventilatory support, and surgical techniques over the past decades. Survival can even be achieved in low-birth-weight infants and mortality is currently limited

to those with coexistent severe life-threatening anomalies [7].

The main complications include a narrow anastomosis, anastomotic strictures, leaks, and recurrent fistula formation. Different factors are responsible for these poor outcomes [8].

Patients and methods

In this prospective case series study, 26 neonates were included, in 2015 and 2016, in the Pediatric Surgery Department, Zagazig University. 11 (42.3%) of these neonates were females and 15 (57.7%) were males

All patients were admitted to NICU and prepared for surgery. 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 cases 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.

Results

	Age (days)	Operative time (min)	CRP	Weight (g)	Hospital stay (days)
Mean	5.4615	62.0769	32.0769	2834.6154	8.69
Median	4.0000	65.0000	12.0000	2900.0000	8.00
SD	3.58029	20.71120	48.43133	356.58638	3.234
Minimum	1.00	4.00	3.00	2150.00	6
Maximum	15.00	100.00	220.00	3400.00	20

Sex distribution

	Outcome		Total	χ^2	*P*
	Lost	Pass			
Sex					
Female					
Count	1	10	11	1.26	0.26
Percentage	20.0	47.6	42.3		
Male					
Count	4	11	15		
Percentage	80.0	52.4	57.7		
Total					
Count	5	21	26		
Percentage	100.0	100.0	100.0		

Age distribution

	Outcome	*N*	Mean	SD	*P*
Age/day	Pass	21	4.3333	2.59487	0.00**
	Lost	5	10.2000	3.42053	

Operation characters

Extubation time	Outcome		Total	χ^2	*P*
	Lost	Pass			
1					
Count	1	3	4	21.7	0.00**
Percentage of outcome	20.0	14.3	15.4		
2					
Count	0	4	4		
Percentage of outcome	0.0	19.0	15.4		
3					
Count	0	2	2		
Percentage of outcome	0.0	9.5	7.7		
Immediate					
Count	0	12	12		
Percentage of outcome	0.0	57.1	46.2		
No					
Count	4	0	4		
Percentage of outcome	80.0	0.0	15.4		
Leakage					
No					
Count	2	16	18	2.48	0.115
Percentage of outcome	40.0	76.2	69.2		
Yes					
Count	3	5	8		
Percentage of outcome	60.0	23.8	30.8		
Total					
Count	5	21	26		
Percentage of outcome	100.0	100.0	100.0		

Associated anomalies×outcome

Associated anomalies	Outcome		Total	χ^2	*P*
	Lost	Pass			
ASD					
Count	0	3	3	14.7	0.04*
Percentage	0.0	14.3	11.5		
Fallot					
Count	1	0	1		
Percentage	20.0	0.0	3.8		
Fallot+limb					
Count	1	0	1		
Percentage	20.0	0.0	3.8		

(***Continued***)

Associated anomalies	Outcome		Total	χ^2	*P*
	Lost	Pass			
No anomalies					
Count	2	14	16		
Percentage	40.0	66.7	61.5		
Syndactyle+hypospadias					
Countus	1	0	1		
Percentage	20.0	0.0	3.8		
Undescended testis					
Count	0	1	1		
Percentage	0.0	4.8	3.8		
Vertebral anomalies					
Count	0	1	1		
Percentage	0.0	4.8	3.8		
VSD					
Count	0	2	2		
Percentage	0.0	9.5	7.7		
Total					
Count	5	21	26		
Percentage	100.0	100.0	100.0		

Comparison

	Outcome	*N*	Mean	SD	*P*
Weight	Pass	21	2911.9048	313.41058	0.02*
	Lost	5	2510.0000	374.83330	
CRP (preoperative)	Pass	21	13.0476	8.99307	0.00**
	Lost	5	112.0000	54.57554	
Operative time	Pass	21	60.4762	15.07521	0.430
	Lost	5	68.8000	34.40182	
Hospital stay	Pass	21	7.38	1.203	0.00**
	Lost	5	14.20	3.347	

*Significant. **Highly significant.

Discussion

EA/TEF is considered one of most challenging pediatric surgery emergencies. The outcome of surgical repair can be evaluated immediately post-operatively, within days or weeks, in terms of mortality.

The assessment of late outcomes among surviving cases in terms of morbidity through follow-up in childhood, adolescence, and adulthood, with a focus on gastro-intestinal-tract (GIT), respiratory, musculoskeletal-related complications, and quality of life, requires studies of longer durations.

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.

In all cases, we first performed repair by open thoracotomy through the third or the fourth space and retropleural dissection to keep the pleural cavity is not opened so, If anastomotic leakage occurred it would be away from pleural cavity.

Preservation of the azygous vein was performed intentionally to preserve the normal anatomy.

Immediate extubation was performed in 12 neonates, with favorable results, to avoid ventilation-associated complications (pneumothorax, pneumonia, and unintentional injuries caused by ventilator artifacts).

In this series, the survival rate was about 80%, which is considered low in comparison with other series.

Rintala *et al*. [9] 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.

Infants undergoing repair of EA and TEF by open or thoracoscopic techniques have excellent survival rates, although favorable outcomes with thoracoscopic repair have been reported in recent reviews comparing the two techniques [10].

Causes of higher mortality in our study in comparison to other studies are the delay before arrival of cases to our hospital, the long period taken by the anesthesiologists to prepare cases preoperatively and the limited number of incubators in our department so cases should wait for a time till there is an empty incubator.

The mean preoperative CRP is 112, which is inversely related to a good outcome; it also contributes to the relatively high mortality.

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].

The azygos vein is the vein that drains the thoracic wall and the upper lumbar region. It is the important channel connecting the two venae cavae. Knowledge of different types of variations is of utmost importance during any surgical intervention and radiological investigation to avoid iatrogenic complications [13].

Ligation division of the fistula and esophago-esophageal anastomosis wasn't difficult with the continuity of the azygous vein.

Conclusion

Open surgical repair of EA, TEF through the retropleural approach with preservation of the azygous vein is feasible and yields good early outcomes, with low mortality, provided that the diagnosis is made and the operation is performed soon also when the birth weight is accepted, CRP level is low and short operation time.

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Conflicts of interest

There are no conflicts of interest.

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