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Original Research Article

Clinical profile of new borns with trachea-esophageal fistula and esophageal atresia and factors associated with outcome at a tertiary care centre

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ABSTRACT

Background: Although trachea-esophageal fistula (TEF) and esophageal atresia (EA) is rare, it is one of the most common congenital anomaly. There is paucity of data due to rarity of condition. More and more data on clinical profile, complications, and outcome are needed to guide the future research.

Objective: To study clinical profile of newborns with TEF and EA and factors associated with outcome.

Materials and Methods: Hospital based prospective study was carried out among 21 newborns with TEF and EA. Diagnosis was confirmed by investigations like X-ray chest after passing nasogastric catheter. Depending upon final diagnosis type of surgery required was determined and procedure was carried out as per the standard guidelines and operating techniques. All cases were followed till the complete post-operative period and the complications and outcome was assessed. Outcome was classified as death or discharge.

Results: Males outnumbered females giving a male to female ratio of 2.5:1. 57.1% did not require ventilator pre-operatively. 61.9% of the cases had no associated abnormality. The most common type of surgery required and performed was tracheoesophageal repair in 52.4% of the cases. Two cases (9.5%) died during the post-operative period. 14 cases (66.7%) recovered and were discharged. Most common complication was anastomotic leak (57.1%). Term status and associated abnormalities were not found to be associated with outcome like death ($p>0.05$).

Conclusion: Trachea-esophageal fistula and esophageal atresia was more common in males. Tracheoesophageal repair was commonly required surgery. Recovery rate was good. Anastomotic leak was most common complication. Outcome like death was not associated with term status and associated abnormalities.

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1. Introduction

In 1697, esophageal atresia (EA) was first described that was associated with trachea-esophageal fistula (TEF) by Thomas Gibson. First surgical repair was performed successfully by Cameron Haight in 1941. EA with or without TEF is one of the most common congenital anomaly. It is a rare condition but it presents challenge for pediatric surgery. In developing countries, this condition is characterized by pneumonitis as

the patients report late to the hospitals.^{1,2}

It has been estimated that esophageal atresia-tracheoesophageal (EA-TEF) incidence is one in 2500-4000 live births.³ It is sporadic in nature. The incidence is more in twin births of about 2.56 times more compared to single births.³

During 4-5 weeks of embryonic development, the there is abnormal septation of caudal foregut leads to TEF. In case the tracheoesophageal septum is positioned abnormally posteriorly, then the trachea and esophagus get connected with each other resulting in EA with fistula.

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If the esophagus cannot recanalize during eighth week of embryonic development, then EA occurs without TEF.⁴

TEF can be suspected in cases of polyhydramnios and also if the stomach bubble is absent in the fetus. Prenatal scans for the diagnosis of EA has low sensitivity. Karyotyping can help in suspected cases.⁵ EA can be diagnosed if it is not possible to pass the nasogastric catheter beyond 10-15 cm in the stomach. X-ray chest will show curled nasogastric catheter in upper esophageal pouch.⁶

TEF is curable and with surgery more than 90% cases survive and hence now a days there is more emphasis on improving quality of life of those who respond to surgery.⁷

There is good prognosis for isolated TEF but cases with both EA and TEF have guarded prognosis and it depends upon the associated abnormalities.⁸

In one study it was found that the esophageal stricture was the most common complication in 35% of cases followed by anastomotic leak in 16% and recurrent fistulae was seen in 3% of the cases.⁹

Although EA-TEF is a rare condition, it is one of the most common congenital anomaly. It is treatable. There is paucity of data on this subject matter due to rarity of the condition. More and more data on clinical profile, complications, and outcome are needed to guide the future research. Hence present study was carried out to study the clinical profile of newborns with trachea-esophageal fistula and esophageal atresia and factors associated with outcome at a tertiary care centre.

2. Materials and Methods

2.1. Study design

Hospital based prospective study.

2.2. Study period

From July 2018 to December 2019.

2.3. Sample size

During the study period, it was possible to include 21 cases of newborns with trachea-esophageal fistula and esophageal atresia as the condition is rare.

2.4. Ethical considerations

Institution Ethics Committee permission was obtained. Child assent was obtained from parents. All cases were managed as per the standard guidelines.

2.5. Settings

Present study was carried out at neonatal intensive care unit, Indira Gandhi institute of child health.

2.6. Inclusion criteria

1. Newborns with tracheoesophageal fistula and esophageal atresia of either gender.
2. Getting admitted and operated at the study site.

2.7. Exclusion criteria

1. Parents not willing to include newborn data with anonymity.
2. Lost to follow up.

2.8. Methodology

Newborns presenting to the Department of Pediatrics, Indira Gandhi institute of child health suspected of having TEF or EA and parents willing to share the data in the present study were included. The diagnosis was confirmed by taking detailed history and investigations like X-ray chest after passing nasogastric catheter.

Baseline data like age, sex, whether pre-term or term baby, gestational age was recorded in the pre designed, pre tested, and semi structured study questionnaire. They were admitted in the Pediatrics wards. Depending upon the requirement, the newborns were kept on either ventilator or were given the mechanical ventilation prior to the surgery to stabilize them. 2-D echo was carried out to find out the associated cardiac abnormalities. All babies underwent investigations like complete blood count, C reactive protein, X-ray, serum electrolytes, arterial blood gases, and blood culture.

Once the baseline pre anesthetic requirements were met with, the newborn was taken for surgery. Depending upon the final diagnosis the type of surgery required was determined and the procedure was carried out as per the standard guidelines and operating techniques. Various surgical procedures performed were End to end anastomosis, Esophageal anastomosis, Esophagostomy, Esophagostomy + gastrostomy, Feeding gastrostomy, Fistula ligation, Gastrostomy, Tracheoesophageal ligation, Tracheoesophageal repair, Thoracotomy + gastrostomy + esophagostomy.

Adequate post-operative care was taken. The child was examined thoroughly. All cases were followed till the complete post-operative period and the complications and outcome was assessed. Outcome was classified as death or discharge.

2.9. Statistical analysis

The data was entered in the Microsoft Excel worksheet and analyzed using proportions. Yates corrected chi square was used and two tailed p value <0.05 was taken as statistically significant.

Table 1: Distribution of study subjects as per baseline parameters

Variable		Number	Percentage
Sex	Male	15	71.4
	Female	06	18.6
Term status	Term baby	18	85.7
	Pre-term baby	03	14.3
	Appropriate for gestational age	16	76.2
Gestational age	Small for gestational age	03	14.3
	Intra uterine growth retardation	02	9.5
Diagnosis	Tracheoesophageal fistula	19	90.5
	Esophageal atresia	02	9.5

3. Results

Males outnumbered females giving a male to female ratio of 2.5:1. There were only 3 (14.3%) pre-term babies. Majority i.e. 16 (76.2%) were appropriate for gestational age. Only two cases (9.5%) were found to have esophageal atresia. (Table 1)

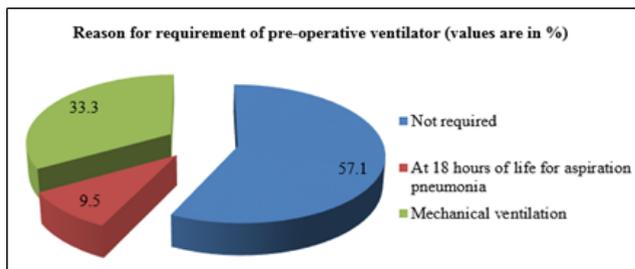


Fig. 1: Distribution of study subjects as per requirement of pre-operative ventilator.

Majority (57.1%) did not require ventilator pre-operatively. Remaining nine cases required it and the most common requirement was mechanical ventilation in 33.3% of the cases. (Figure 1)

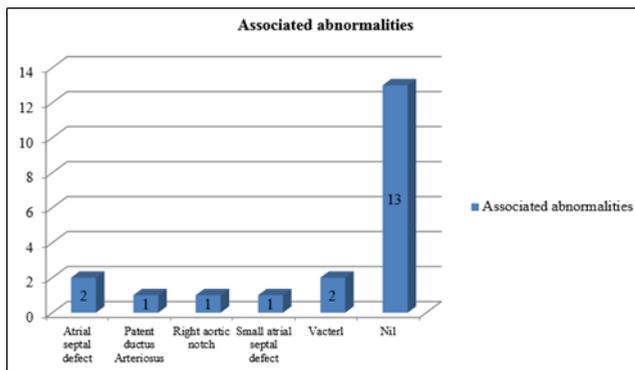


Fig. 2: Distribution of study subjects as per associated abnormalities.

Majority (61.9%) of the cases had no associated abnormality. Two cases had associated atrial septal defect (9.5%). Vacterl was seen in only two cases (Figure 2)

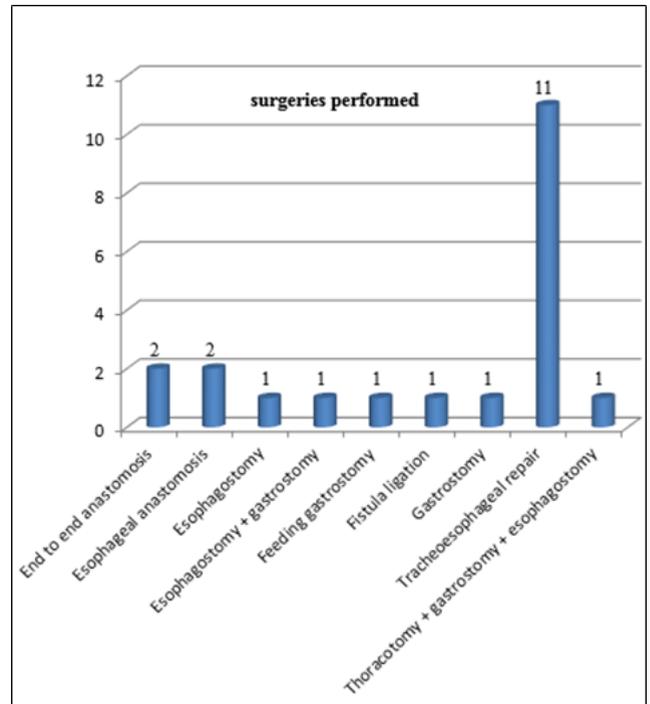


Fig. 3: Distribution of study subjects as per surgery performed

The most common type of surgery required and performed was tracheoesophageal repair in 52.4% of the cases. (Figure 3)

Five cases (23.8%) got Discharge against medical advice while two cases (9.5%) died during the post-operative period. 14 cases (66.7%) recovered and were discharged (Figure 4)

Most of the cases (57.1%) had Anastomotic leak. Among others was Gastro esophageal reflux disease in three cases (14.3%) followed by stricture in two cases and two cases had no complications (Table 2)

We examined an association between term status and outcome. There were two deaths in term babies compared to zero in pre term babies but this difference was not found to be statistically significant (p>0.05). (Table 3)

Table 2: Distribution of study subjects as per post-operative complications

Post-operative complications	Number	Percentage
Nil	02	9.5
Anastomotic leak	12	57.1
Gastro esophageal reflux disease	03	14.3
Respiratory morbidity	01	4.8
Stricture	02	9.5
Tracheomalacia	01	4.8
Total	21	100

Table 3: Association between term status and outcome

Term status	Outcome			Total	Chi square	P value
	Death	Discharge	DAMA			
Pre term	0	2 (9.5%)	1 (4.8%)	3 (14.3%)	0.4667	0.792
Term	2 (9.5%)	12 (57.1%)	4 (16.1%)	18 (85.7%)		
Total	2 (9.5%)	14 (66.7%)	5 (23.8%)	21 (100%)		

Table 4: Association between associated abnormalities with outcome

Associated abnormalities	Outcome			Total	Chi square	P value
	Death	Discharge	DAMA			
No	2 (9.5%)	8 (38.1%)	3 (14.3%)	13 (61.9%)	1.988	0.369
Yes	0	7 (33.3%)	1 (4.8%)	8 (38.1%)		
Total	2 (9.5%)	15 (71.4%)	4 (19.1%)	21 (100%)		

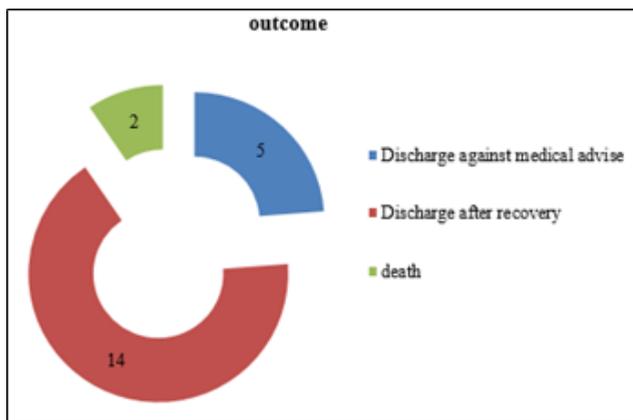


Fig. 4: Distribution of study subjects as per the outcome

Among Babies with no associated abnormalities two deaths were encountered compared to nil among those with associated abnormalities. But this difference was not found to be statistically significant ($p>0.05$). (Table 4)

4. Discussion

Males outnumbered females giving a male to female ratio of 2.5:1 in the present study. Al-Salem AH et al¹⁰ in their review of 94 cases also observed that males were than females (55 vs. 39). Tsai JY et al¹¹ in their analysis of 81 cases over two decades also noted that males outnumbered over females (46 vs. 35). Engum SA et al¹² also reported

male preponderance from their study (127 vs. 100). Acher CW et al¹³ also noted that 56% in their study subjects were males.

There were only 3 (14.3%) pre-term babies. Majority i.e. 16 (76.2%) were appropriate for gestational age. Only two cases (9.5%) were found to have esophageal atresia.

Majority (57.1%) did not require ventilator pre-operatively. Remaining nine cases required it and the most common requirement was mechanical ventilation in 33.3% of the cases. There were two cases who had aspiration pneumonia while Al-Salem AH et al¹⁰ in their study found that the incidence of aspiration pneumonia was very high i.e. 39.4% at the time of admission.

Majority (61.9%) of the cases had no associated abnormality. Two cases had associated atrial septal defect (9.5%). Vacterl was seen in only two cases. AH et al¹⁰ in their study found that 49% had associated abnormalities which are higher than the present study of 38.1%. Engum SA et al¹² had reported from their study that 64% of the babies had associated abnormalities. Friedmacher F et al¹⁴ reported from their study that 64.2% had associated abnormalities.

The most common type of surgery required and performed was tracheoesophageal repair in 52.4% of the cases. Five cases (23.8%) got Discharge against medical advice while two cases (9.5%) died during the post-operative period. 14 cases (66.7%) recovered and was discharged. The mortality rate was high (30.8%) in the study carried out by AH et al.¹⁰ In the study by Tsai JY et al¹¹ the most common surgery performed was primary

esophageal anastomosis in 44 cases out of total 81 cases included. Tsai JY et al¹¹ found that the mortality rate was 22% which is higher than the present study of 9.5%. Engum SA et al¹² had performed a single-layer anastomosis 81% of the cases. In their study the mortality rate was only 5% which is lower than the present study. Friedmacher F et al¹⁴ also carried out primary surgical repair in majority of their study subjects and the death rate was only 4.2% during the post operative period. Acher CW et al¹³ also observed in their study that standard open repair was most commonly performed surgery.

Most of the cases (57.1%) had Anastomotic leak. Among others was Gastro esophageal reflux disease in three cases (14.3%) followed by stricture in two cases and two cases had no complications. In the study by Tsai JY et al¹¹ the most common complication was stricture in 40% of the cases followed by anastomotic leak in 19% of the cases among those who underwent primary esophageal anastomosis. Engum SA et al¹² had reported from their study that 16% had anastomotic leak, 35% had stricture, 3% had recurrent fistula. Incidence of post operative leak was 26% in the study by Acher CW et al.¹³

We examined an association between term status and outcome. There were two deaths in term babies compared to zero in pre term babies but this difference was not found to be statistically significant ($p>0.05$). Among Babies with no associated abnormalities two deaths were encountered compared to nil among those with associated abnormalities. But this difference was not found to be statistically significant ($p>0.05$). But Tsai JY et al¹¹ found that the death rate was more in those with the associated abnormalities.

5. Conclusion

Trachea-esophageal fistula and esophageal atresia was more common in males. Tracheoesophageal repair was commonly required surgery. Recovery rate was good. Anastomotic leak was most common complication. Outcome like death was not associated with term status and associated abnormalities.

6. Conflict of Interest

The authors declare that there are no conflicts of interest in this paper.

7. Source of Funding

None.

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