Original Article

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Analysis of clinical characteristics and comparison of survival of inborn versus outborn cases of esophageal atresia with tracheo-esophageal fistula

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KEYWORDS

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ABSTRACT

Background: Many factors influence the survival of newborns with esophageal atresia and tracheoesophageal fistula, including birth weight, maturity at birth, and associated congenital anomalies. However, the impact of the place of delivery on the survival of esophageal atresia cases has rarely been explored. This study aims to compare the survival rates of newborns with esophageal atresia and tracheoesophageal fistula (EA/TEF) born at our tertiary care center (inborn) with those born elsewhere and referred to our center for management (outborn).

Methods: This retrospective observational study was conducted from June 2021 to December 2023 at a tertiary care center in North India. A total of 46 cases of EA/TEF were included. These cases were divided into two groups: outborn and inborn. Outborn cases comprised those delivered at other hospitals and admitted through the pediatric emergency department of our institute. Inborn cases were delivered at our institute and admitted to the neonatal ICU managed by neonatologists.

Results: Among the 46 cases, 17 (36.96%) were inborn, and 29 (63.04%) were outborn. The average birth weight in the inborn group was 1.74 ± 0.53 kg, compared to 2.22 ± 0.34 kg in the outborn group ($p = 0.0006$). Preterm births were more common in the inborn group than in the outborn group ($p = 0.004$). Associated VACTERL anomalies were present in 6 cases in each group, accounting for 35.29% of inborn cases, 20.69% of outborn cases, and 26.09% overall. The overall survival rate was 41.30%. Survival was higher in the inborn group (58.82%) compared to the outborn group (31.03%), though this difference was not statistically significant ($p = 0.064$).

Conclusion: The survival of newborns with esophageal atresia in developing nations depends on various factors beyond birth weight, maturity, and associated anomalies. The availability of pediatric surgical facilities and neonatology support at the place of delivery significantly impacts outcomes and warrants further detailed study.

INTRODUCTION

Esophageal atresia (EA), with or without tracheoesophageal fistula (TEF), is a rare congenital anomaly occurring in 1–2 per 5,000 live births. [1,2] It is characterized by a congenital disruption in the continuity of the esophagus, with or without abnormal communication to the trachea. Gross classified EA into five types based on the presence and nature of this tracheal communication. [3] The most common variant, Type C (86%), features a blind-ending upper esophageal pouch and a lower esophageal pouch that communicates with the trachea. [3,4]Antenatal diagnosis of EA is rare because the distal communication with the trachea enables swallowing of amniotic fluid, thereby preventing the maternal polyhydramnios often associated with such anomalies. [5,6] As a result, the diagnosis is typically made postnatally, based on clinical findings such as excessive salivation since birth, dyspnea, choking, and radiological evidence of nasogastric tube coiling in the superior mediastinum (T2–T4) on chest X-ray. [7]

Survival in EA/TEF depends on several factors, including the presence of associated cardiac anomalies, gestational maturity, and birth weight. [8] In developing nations like India, where access to specialized pediatric surgical care is limited, timely diagnosis and management of EA/TEF pose

significant challenges. Newborns are often delivered at centers lacking such facilities, resulting in delayed referrals. Furthermore, inadequate transport practices—such as failure to maintain normothermia, oxygenation, and proper suctioning—further contribute to the mortality of these newborns.

This retrospective observational study aims to compare the clinical characteristics and survival rates of inborn cases of EA/TEF (born and treated at our tertiary care center) with outborn cases (born at other hospitals and referred to our center for surgical management).

METHODS

This retrospective, single-center observational study was conducted from June 2021 to December 2023 at a tertiary care center in North India, a region with a high prevalence of congenital malformations. A total of 46 cases of esophageal atresia with tracheoesophageal fistula (EA with TEF) were included. Data were retrieved from patient admission records.

Study Groups

The cases were divided into two groups:

Outborn: Delivered at other hospitals and admitted through the pediatric emergency department.

Inborn: Delivered at the study institute and managed in the neonatal ICU by neonatologists.

Inclusion Criteria: Operated cases of EA with TEF.

Exclusion Criteria: Cases of pure esophageal atresia without fistula.

Preoperative Management

Upon admission, all cases were clinically examined and medically stabilized with:

Continuous oral and nasal suction; Adequate warming; and Intravenous fluids.

Diagnostic investigations included chest and abdominal radiographs with a wide-bore feeding tube in situ to confirm coiling and assess the level of coiling, as well as the presence of abdominal gas shadow. Screening echocardiograms and renal ultrasounds were performed to identify cardiac and renal anomalies, though logistical challenges often limited their use due to the risks associated with transporting neonates.

Operative Procedure

Diagnosis-confirmed cases underwent surgical repair through a standard right posterolateral thoracotomy. Key steps included:

- Identification of the upper and lower esophageal pouches.
- Measurement of the inter-pouch gap using a sterile measuring scale.
- Transfixation and division of the tracheoesophageal fistula near the tracheal opening.
- Mobilization of the upper pouch to achieve tension-free anastomosis using interrupted 5-0 absorbable polyglactin sutures over an 8-Fr transanastomotic feeding tube.
- In cases where anastomosis was not feasible, cervical esophagostomy and gastrostomy were performed.
- Closure of the thoracic incision with the placement of an intercostal drainage tube.

Postoperative care was conducted in neonatal/pediatric surgery ICUs.

Parameters Assessed

The following variables were analyzed:

- Maturity and birth weight.
- Age at surgery.
- Sex.
- Associated anomalies.
- Intraoperative gap between esophageal pouches.
- Postoperative status (intubated or extubated).
- Type of surgical procedure.
- Survival.

Statistical Analysis

Data were analyzed using SPSS-PC version 25 (Chicago, Illinois). Quantitative data (nonparametric) were expressed as mean ± standard deviation or median with interquartile range. Qualitative data were expressed as frequencies and percentages. Chi-square or Fisher's exact test was used to compare proportions. A p-value <0.05 was considered statistically significant.

RESULTS

A total of 46 cases of esophageal atresia with tracheoesophageal fistula (EA with TEF) were included in this study. Two cases of pure esophageal atresia were excluded. Out of these 46 cases, 17 (36.96%) were inborn, and 29 (63.04%) were outborn. The overall male-to-female ratio was 1.09:1 (24:22).

In terms of maturity, preterm babies were more prevalent in the inborn group compared to the outborn group, with a P-value of .004. When birth weight was compared, there was a statistically significant difference between the inborn and outborn groups, with 7 newborns below 1.5 kg (very low birth

weight) in the inborn group and none in the outborn group. The average birth weight in the inborn group was 1.74 ± 0.53 kg compared to 2.22 ± 0.34 kg in the outborn group, with a P-value of .0006 [Table 1].

Table 1: Comparison of clinical characteristics of Inborn and Outborn group of patients

PARAMETERS		**INBORN (n=17)**	**OUTBORN (n=29)**	**TOTAL (N=46)**	***P value***
MATURITY	Preterm	9 (52.94%)	4 (13.79%)	13 (28.26%)	.004
	Term	8 (47.06%)	25 (86.21%)	33 (71.74%)	
BIRTH WEIGHT (Kg)	≥2.5	1 (5.88%)	6 (20.69%)	7 (15.22%)	.0006
	1.5-2.4	9 (52.94%)	23 (79.31%)	32 (69.56%)	
	<1.5	7 (41.18%)	0 (0.00%)	7 (15.22%)	
AGE AT SURGERY (days)	2-3	11 (64.70%)	19 (65.52%)	30 (65.22%)	.453
	4-5	3 (17.65%)	8 (27.57%)	11 (23.91%)	
	>5	3 (17.65%)	2 (6.91%)	5 (10.87%)	
SEX	Female	7 (41.18%)	15 (51.72%)	22 (47.83%)	.489
	Male	10 (58.82%)	14 (48.28%)	24 (52.17)	
ASSOCIATED VACTERL ANOMALIES	Cardiac	1	3	4	
	Anorectal	1	3	4	
	Renal	1	nil	1	
	Bowel atresia	1	nil	1	
	Limb anomalies and others	2	nil	2	
GAP (cm)	≤2	10(58.82%)	19(65.52%)	29(63.04%)	.092
	2-3	3(17.65%)	9(31.03%)	12(26.09%)	
	>3	4(23.53%)	1(3.45%)	5(10.87%)	
POSTOPERATIVE STATE	Intubated	16(94.12%)	19(65.52%)	35(76.09%)	.027
	Extubated	1(5.88%)	10(34.48%)	11(23.91%)	
PROCEDURE	Primary esophageal anastomosis	13(76.47%)	23(79.31%)	36(78.26%)	.824
	Esophagostomy and gastrostomy	4(23.53%)	6(20.68%)	10(21.74%)	
OUTCOME	SURVIVED	10(58.82%)	9(31.03%)	19(41.30%)	.064
	DIED	7(41.18%)	20(68.97%)	27(58.70%)	

The median age at surgery in both the inborn and outborn groups was 3 days, with no statistically significant difference. Associated components of VACTERL anomalies were observed in 6 cases in each group (35.29% in the inborn group and 20.69% in the outborn group, 26.09% overall), with no statistically significant difference [Table 1].

Intraoperatively, the gap between the upper and lower pouches was studied in both groups. A gap < 2 cm was noted in 58.82% of cases in the inborn group and 65.52% of cases in the outborn group. A gap > 3 cm was observed in 23.53% of cases in the inborn group and 3.45% of cases in the outborn group. However, there was no statistically significant difference between the two groups (P = .092). Primary esophageal anastomosis was performed in 76.47% of cases in the inborn group and 79.31% of cases in the outborn group, with no statistically significant difference [Table 1].

When the postoperative status was analyzed in the two groups in terms of intubation or extubation, more cases remained intubated in the immediate postoperative period in the inborn group (94.12%) compared to the outborn group (65.52%), with a statistically significant difference (P = .027) [Table 1].

When the cases were classified according to the Waterston classification, the majority of cases in the inborn group were type C (58.82%), whereas in the outborn group, the majority were type B (68.97%), which was statistically significant (P = .05) [Table 2].

Overall survival was 41.30%. Survival was higher in the inborn group (58.82%) compared to the outborn group (31.03%). However, this was not statistically significant (P = .064). The relationship of other parameters with survival was analyzed separately. It was found that the survival of both term and preterm neonates was higher in the inborn group compared to the outborn group, with a P-value of .033. However, for other parameters, there was no statistically significant difference [Table 3]. Survival was also studied based on the Waterston classification. It was noted that in the inborn group, survival was 100% in type A, 83.33% in type B, and 40% in type C. This observation is comparable to the original Waterston classification in terms of survival rates. In the outborn group, survival was 45% in type B and 0% in type C. There was a statistically significant difference between the inborn and outborn groups when survival based on the Waterston classification was compared, with a P-value of 0.048 [Table 2].

Table 2: Comparison of Inborn Vs Outborn group in terms of Waterston classification

Waterston type	**INBORN**		**OUTBORN**		**TOTAL**		***P-Value***	
	Case distribution (n=17)	**Survival**	**Case distribution (n=29)**	**Survival**	**Case distribution (n=46)**	**Survival**	**Case distribution**	**Survival**
A (>2.5Kg and well)	1(5.88%)	1(100%)	0(0.00%)	--	1(2.7%)	1(100%)	.05	.048
B(1.8-2.5Kg , or >2.5kg with pneumonia and congenital anomalies	6(35.3%)	5(83.33%)	20(68.97%)	9(45%)	26(56.52%)	14(53.8%)		
C(<1.8Kg, or with severe pneumonia or congenital anomalies)	10(58.82%)	4(40%)	9(31.03%)	0(0%)	19(41.30%)	4(21%)		

Table 3: Comparison of survival in terms of various parameters in Inborn Vs Outborn group

PARAMETERS		**SURVIVAL(%age)**			***P- Value***
		INBORN (n=17)	**OUTBORN (n=29)**	**TOTAL (N=46)**	
MATURITY	Preterm	4(9; 44.44%)	0(4; 00.00%)	4(13; 30.77%)	.033
	Term	6(8; 75.00%)	9(25; 36.00%)	15(33; 45.45%)	
BIRTH WEIGHT (Kg)	≥2.5	1(1; 100.00%)	1(6; 16.66%)	2(7; 28.57%)	.198
	1.5-2.4	6(9; 66.67%)	8(23; 34.78%)	14(32; 43.75%)	
	<1.5	3(7; 42.86%)	----	3(7; 42.86%)	
AGE AT SURGERY (days)	2-3	4(11; 36.36%)	6(19; 31.58%)	10(30; 33.33%)	.463
	4-5	3(3; 100.00%)	1(8; 12.50%)	4(11; 36.36%)	
	>5	3(3; 100.00%)	2(2; 100.00%)	5(5; 100.00%)	
SEX	Female	5(7; 71.43%)	3(15; 20.00%)	8(22; 36.36%)	.462
	Male	5(10; 50.00%)	6(14; 42.86%)	11(24; 45.83%)	
ASSOCIATED VACTERL ANOMALIES	Cardiac	0(1; 00.00%)	2(3; 66.67%)	2(4; 50.00%)	--
	Anorectal	0(1; 00.00%)	0(3; 00.00%)	0(4; 00.00%)	
	Renal	1(1; 100.00%)	-	1(1; 100.00%)	
	Bowel atresia	0(1; 00.00%)	-	0(1; 00.00%)	
	Limb anomalies and others	0(2; 00.00%)	-	0(2; 00.00%)	
GAP (cm)	≤2	8(10; 80.00%)	7(19; 36.84%)	15(29; 51.72%)	.505
	2-3	2(3; 66.67%)	1(9; 11.11%)	3(12; 25.00%)	
	>3	0(4; 00.00%)	1(1; 100.00%)	1(5; 20.00%)	
POSTOPERATIVE	Intubated	9(16; 56.25%)	5(19; 26.31%)	14(35; 40.00%)	.32

STATE	Extubated	1(1; 100.00%)	4(10; 40.00%)	5(11; 45.45%)	
PROCEDURE	Primary esophageal anastomosis	10(13; 76.92%)	9(23; 39.13%)	19(36; 52.78%)	--
	Esophagostomy and gastrostomy	0(4; 00.00%)	0(6; 00.00%)	0(10; 00.00%)	

DISCUSSION

Esophageal atresia is a rare congenital anomaly in newborns, with various factors contributing to survival. [9-11] In developing countries like India, where the majority of the population is still under low socioeconomic status, the incidence of congenital anomalies such as EA is relatively higher compared to developed nations. [12, 13] Simultaneously, the healthcare infrastructure and availability of pediatric surgical facilities and expert neonatal care are still deficient in remote areas, making early diagnosis and management challenging. This often leads to delayed referrals to higher centers, which are associated with suboptimal care during transportation, such as failure to maintain euthermia and inadequate oronasal suction, resulting in aspiration pneumonitis. [14] By the time a newborn reaches a center equipped with pediatric surgical facilities, their general condition is often compromised, leading to higher mortality rates.

On the contrary, when a newborn with EA is delivered at a center with pediatric surgical and neonatal ICU facilities, immediate postnatal diagnosis and management—including the maintenance of euthermia, frequent oronasal suction, intubation as required, and hemodynamic stabilization—followed by timely surgical repair, significantly improve survival rates. Although many factors affecting outcomes in these babies have been studied, there is limited literature comparing survival rates in inborn versus outborn cases. One study by Schlee D et al. concluded that there is no significant difference in outcomes between inborn and outborn cases of EA. [15] However, this study originates from the western world, where the incidence is low and healthcare infrastructure is of high quality. There is a paucity of such literature from developing nations. Hence, our study provides insight into this comparison and serves as a foundation for further detailed studies.

In this single-center retrospective observational study, we analyzed and compared the clinical characteristics and survival rates of EA with TEF cases born in our center (inborn) with those born elsewhere and referred to our center for management (outborn).

Out of the 46 patients with EA and TEF included in the study, 17 (36.96%) were inborn, and 29 (63.04%) were outborn. This finding is comparable to a study by Seo J et al., in which 38% of cases were inborn. [16] Preterm newborns with EA and TEF predominated in the inborn group (9 cases, 52.94%) compared to the outborn group (4 cases, 13.79%), with a statistically significant P-value of 0.004. Of these preterm newborns, 4 (44.44%) survived in the inborn group, whereas none survived in the outborn group, with a P-value of 0.03, which is statistically significant. This highlights that delivery at a center with pediatric surgical and advanced neonatal facilities is a crucial factor in the outcomes of EA and TEF cases. For term babies as well, survival was higher in the inborn group (75%) compared to the outborn group (36%), further supporting this observation.

The cases were also classified according to the Waterston classification in both groups. When survival based on the Waterston classification was compared between the two groups, the survival pattern in the inborn group aligned with the original Waterston classification [17], whereas survival was significantly lower in the outborn group, with a P-value of 0.048. This finding further underscores the favorable outcomes for inborn newborns with EA compared to outborn patients.

The median age at surgery was 3 days in both groups. However, 3 patients in the inborn group and 2 patients in the outborn group underwent surgery after 5 days. In the outborn group, the delay was due to late presentation. In the inborn group, one patient was a preterm (32 weeks' gestation) with a birth weight of 1.2 kg, requiring initial resuscitation and stabilization before being deemed fit for anesthesia. Another newborn, born to an Rh-negative mother, required two cycles of exchange blood transfusion, causing a delay in surgical readiness. The third was a term baby with aspiration pneumonitis who required intubation and stabilization before surgery. Importantly, all cases operated after 5 days survived, a finding that warrants further evaluation. This is noteworthy because most literature recommends early surgery for EA, preferably within 48 hours of life, for optimal outcomes. [18]

The immediate postoperative status is also a significant factor. In the inborn group, 9 out of 16 cases (56.25%) kept intubated postoperatively survived, compared to only 5 out of 19 cases (26.31%) in the outborn group. This suggests more severe preoperative respiratory damage in the outborn cases, contributing to poor outcomes, though the difference was not statistically significant.

Evaluation of associated anomalies to rule out VACTERL is a prerequisite in managing EA cases, as 20-30% of cases are associated with such anomalies. [19] In our study, anorectal malformations and musculoskeletal anomalies were assessed in all cases using clinical examination and plain radiographs, which are readily available. However, echocardiograms and renal ultrasonography, essential for detecting cardiac and renal anomalies, respectively, could not be performed for all cases due to logistical limitations (non-availability of bedside facilities). Cardiac pathology was detected in 4 cases (1 inborn, 3 outborn), and 1 renal anomaly (multicystic dysplastic kidney) was identified in the inborn group based on antenatal ultrasound findings.

Overall, survival was higher in the inborn group (58.82%) compared to the outborn group (31.03%). The survival rate in the inborn group closely matches the survival rates reported in the literature from developing nations. [10, 11, 20] This finding emphasizes the importance of providing pediatric surgical and neonatal care facilities at every district-level hospital in countries like India, where congenital malformations are more prevalent compared to developed nations.

This was a retrospective single-center observational study; therefore, the findings cannot be generalized to the entire population. Multicenter studies are needed for broader conclusions. Additionally, proper assessment of associated congenital anomalies was limited by logistical issues, potentially influencing survival rates in the outborn group.

CONCLUSION

The survival of esophageal atresia in developing nations depends on factors beyond birth weight, maturity, and associated anomalies. The availability of pediatric surgical and neonatal care facilities at the place of delivery significantly affects outcomes and warrants further detailed studies. Simultaneously, there is a need to improve the referral system for neonates with congenital malformations like EA, ensuring adequate temperature monitoring, suction, and oxygenation during transport to enhance survival rates.

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