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THE OUTCOME IN TEF PATIENTS PRESENTING AFTER ONE WEEK OF LIFE

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ABSTRACT

Background and aim: Esophageal atresia (EA) is a rare congenital malformation consisting of a lack of continuity between the upper and lower esophageal pouches, frequently associated with tracheoesophageal fistula which occurs in 1/2500 to 1/4000 live births. In developing countries late presentation of patients with Esophageal Atresia (EA) can be multifactorial which includes difficulty in access to health care facilities and less awareness among primary level health care workers. Present case series presented to study outcome and factors associated with EA among patients presented after 7 days of live birth.

Methodology: All neonates with EA admitted in the Neonatal Intensive Care Unit (NICU) of the department of paediatric surgery were included in the study. Patients presented after 7 days of life during September 2018 to November 2019 were included in the study. The data regarding clinical history, general and systemic examination, pre-operative investigations, intraoperative findings and outcome of surgery were recorded.

Results: Out of all EA patients managed during study period, 18 (5.12%) patients were presented after 7 days. Male to female Ratio was 1:1. Mean age was higher among deceased (14.6±8.2 days) than who survived (10.5±8.2). Mean birth weight was also found lower among deceased (1.97±0.99) than survived (2.10±1.03). Associated congenital anomalies were present in 15 (83.3%) patients. In 9 (50%) patients more than one anomaly was present. Preoperatively 5 (28%) patients need ventilatory care. Mortality among them was 80%. Post-operative anastomotic leak was present in 3 (16.7%) patients while 15 (83.3%) patients found no major postoperative complication. Mortality was found higher among patients with low birth weight, long gap length, presented with respiratory complication. Statistically no factor was related to outcome.

KEYWORDS :

INTRODUCTION:

Esophageal atresia (EA) is a rare congenital malformation consisting of a lack of continuity between the upper and lower esophageal pouches, frequently associated with tracheoesophageal fistula which occurs in 1/2500 to 1/4000 live births. [1] William Durston had first described congenital atresia of the esophagus in 1670. [2] The survival of neonates improved dramatically once Cameron Haight's reported successful surgical correction of EA in 1941. [3] Advances in neonatal intensive care, anesthetic management, ventilatory support and surgical techniques are major contributory factors in improving survival over the past decades. In 1962 Waterston had reported low birth weight, pneumonia and associated congenital anomalies as the risk factors of EA outcome. Based on that he proposed a prognostic classification for EA [4] In Waterston study mortality was found very high (94%) among group C followed by group B (32%) and group A (6%) Advances in neonatal intensive care, anesthetic management, ventilatory support and surgical techniques are major contributory factors in improving survival over the past decades. Even low birth weight babies can be survived now a day, [5] and the mortality is currently limited to those with coexistent severe life-threatening anomalies.

In developing countries like India reason behind late presentation can be multifactorial. Factors related to public health services like difficulty in access to the health care facility, home deliveries or the less awareness of the condition amongst primary level health care providers may contribute to the late presentation. There are few case reports of neonates who survived after the surgical correction of EF after the late presentation. [6,7] Present case series presented to study the outcome and factors associated with EA.

OBJECTIVES:

- To study the outcome of surgical intervention of Esophageal Atresia (EA) in neonates presented after one week.
- To identify factors associated with outcome.

METHODOLOGY:

The cross-sectional observational study was carried out in the Paediatric Surgery Department of XYZ Hospital, ABC city, Rajasthan.

All neonates with EA admitted in the Neonatal Intensive Care Unit (NICU) of the department of pediatric surgery during September 2018 to November 2019 were included in the study. Patients presented after 7 days of life were included in the study while patients presented within first-week were excluded. Patients transferred to the department after corrective surgery performed outside were excluded from the study.

Detailed clinical history including the presenting symptoms, maternal history, birth history, general and systemic examination was taken and data was collected in predesigned proforma. Also, preoperative investigations undertaken for the establishment of diagnosis, associated anomalies, operative procedure, intraoperative findings, postoperative complications, and treatment outcomes.

Classification of EA was done according to Gross's anatomic classification. The diagnosis of EA was confirmed by radiographs (both anteroposterior and lateral views) with a red rubber catheter in situ. By taking delayed presentation in most of the cases with pneumonia into consideration, chest drain was inserted in all patients with EA undergoing thoracotomy, Intravenous fluids, antibiotics, and parenteral nutrition were continued postoperatively. Follow-up was not done in the study.

RESULT:

Table 1: Mean age, sex and birth weight of patients (N=18)

	Deceased	Survived	P value
Age	14.6±3	10.5±7.6	0.28
Sex			
Male	7	2	1.0
Female	7	2	
Birth weight	1.97 ± 0.99	2.10 ± 1.03	0.198

Out of the 351 patients who were managed for EA, 18 (5.12%) presented after 7 days of life. Male: Female ratio was 1:1. Mean age was higher among deceased (14.6±8.2 days) than who survived (10.5±8.2) and this difference was found statistically significant. Mean birth weight was also found lower among deceased (1.97±0.99) than survived (2.10±1.03) but this difference was found statistically non-significant.

Table 2: Distribution of congenital anomalies among patients with EA (N=18)

	Anomaly	N (%)
A.	**No anomalies**	3 (16.7%)
B.	**ARM**	13 (72.2%)

C.	**Cardiac**	10 (55.6%)
	Dextrocardia	2
	Atrial Septal Defect	2
	RAA	1
	PFO	2
	CAA	1
	Tetralogy of folate	1
	Patent Ductus arteriosus	1
D.	**RENAL and GENITAL**	1 (5.6%)

Associated congenital anomalies were present in 15 (83.3%) patients. In 9 (50%) patients more than one anomaly was present. The details of their characteristics are given in Table 1.

Table 3: Comparison of preoperative parameters among survived and deceased patients

	Parameters	Total	%	Survived (n=4)	%	Deceased (n=14)	%	P value
Presenting complaints	Inability to tolerate feeds	14	78%	3	21%	11	79%	0.87
	Excessive frothing from mouth	18	100%	4	22%	14	78%	NA
	Respiratory distress	4	22%	1	25%	3	75%	0.87
Examination findings	Distended abdomen	8	44%	2	25%	6	75%	0.79
	Scaphoid abdomen	11	61%	2	18%	9	82%	0.6
	Red rubber catheter negotiable beyond 10 cm	3	17%	1	33%	2	67%	0.61
	Perineum: Anal opening	13	72%	3	23%	10	77%	0.88
	Signs of sepsis	6	33%	1	17%	5	83%	0.69
	Cardiac murmur	10	56%	2	20%	8	80%	0.79
Congenital anomalies	With one or more anomalies	15	83%	4	27%	11	73%	NA

All patients were presented with excessive frothing from mouth while inability to tolerate feed was complained by 14 (78%) patients and respiratory distress was there in 4(22%) patients. On examination distended abdomen was present in 8 (44%) patients while scaphoid abdomen was present in 11 (61%) patients. Red rubber catheter negotiable beyond 10 cm in 3 (17%) patients. At the time of admission signs of sepsis was present in 6 patients out of which one patient survived. Table 2 summarize the presenting complaints and examination finding of presented babies.

Signs of sepsis were present in 2 patients. On babygram pneumonitis and consolidation of lung was found in 3 patients. Out of these, 1 patient survived post-operatively. High CRP was found among 4 (22%) patients. Radiologically most commonly found position of red rubber catheter was at T3 level in 8 (44%) patients followed by at T4 in 7 (39%) and in 1 (6%) patient at T2 level. In 11 (61%) patients both pneumonitis and consolidated lung was found and in 1 patient sign of pleural effusion and lung consolidation was found. In 5 patients condition deteriorated initially before surgery. These babies were born at full-term and all of them were of Waterston's type C. Postoperatively leak was found in 3 out of 18 patients. Seven patients were put on nebulizer with epinephrine and 11 patients were kept on albuterol nebulizer. Saline nebulization was given to 7 patients. Out of all patients 2 patients develop minor leak and 1 patient develop major leak. Among all patients presented after 7 days of life 4 (22.22%) patients were survived.

Table 4: Comparison of preoperative parameters among survived and deceased patients

	Parameters	Total	%	Survived (n=4)	%	Deceased (n=14)	%	P value
Preoperative investigations	High CRP	4	22%	1	25%	3	75%	0.87
Babygram	Position of red rubber catheter	0	0%					NA
	T2	1	6%	0	0%	1	100%	
	T3	8	44%	3	38%	5	63%	
	T4	7	39%	1	14%	6	86%	
	H type	2	11%	0	0%	2	100%	
	Distal bowel gas	18	100%	4	22%	14	78%	NA
	Pneumonitis	11	61%	2	18%	9	82%	0.61
	Consolidation of lungs	16	89%	4	25%	12	75%	NA
	Pleural effusion	1	6%	0	0%	1	100%	NA
Preoperative critical care	Put on ventilatory support	5	28%	1	20%	4	80%	0.89
Waterston classification	Waterston A	1	6%	0	0%	1	100%	Na
	Waterston B	4	22%	1	25%	3	75%	
	Waterston C	13	72%	3	23%	10	77%	

Comparison of preoperative variables of patients who survived and those who deceased shown in table 4. Most of the variables do not exhibit significant difference in both groups.

Table 5: Intraoperative gap length and postoperative complication

Gap length	Total	Survived (%)		Deceased (%)		P value
<2 cm	13 (72.2%)	4	30.8%	9	69.2%	0.27
≥ 2 cm	5 (27.8%)	0	0%	5	100%	
Postoperative complication						
Anastomotic leak	3 (16.7%)	0	0%	3	100%	1
No complication	15 (83.3%)	4	26.7%	11	73.3%	

Mortality was found high among patients with high gap length than short gap length but difference was found statistically non-significant. Post-operative anastomotic leak was present in 3 (16.7%) patients while 15 (83.3%) patients were found no major postoperative complication.

DISCUSSION:

In the present study, a group of patients who presented late after 7 days of live birth were included. Out of all patients managed for EA, 18 (5.12%) patients were presented after 7 days. There is variation among studies regarding the proportion of patients presented late. Nagdeve et al [8] also found that 8% of patients presented after the age of 7 days while Gupta et al [9] found only 3% of patients presented after 7 days of birth. The majority of the cases presented late were from Waterston's group B and C. Such late presentation might be due to poverty, lack of awareness, poor health-seeking behavior, lack of accessibility to health care facilities. This also indicates that the diagnosis was missed at an institute or hospital where babies were delivered.

Tandon et al [10] observed male preponderance in these late presenters. While in the present study no differentiation observed among patients presented after 7 days. Associated congenital anomalies were present in 15 patients. In 9 patients more than one anomaly was present. Anatomical aspects of EA in a group of patients with late presentation shown some importance regarding the outcome. Out of all, 5 patients had distance between two pouch of the oesophagus which was 2 cm or more while the remaining 13 patients had a gap less than 2 cm. All patients with a long gap died postoperatively. Postoperatively anastomotic leak was present in 3 patients. Some studies have evidenced that associated congenital anomalies, postoperative complications and mortality are an outcome of gap length. [11,12] With

contrast to the present study, Nagdeve et al found that intolerance to feeding resulted in a statistically worse outcome. In the present study, 78% of patients had an intolerance to feeding.

Pneumonitis and lung consolidation were present in 11 (61%) and 16 (89%) respectively. Out of 14 deceased, 9 patients had pneumonitis and 12 patients had lung consolidation. One-third of patients have presented with sepsis and mortality among them was 83%. Although pneumonia didn't found the association in the present study, some other author pneumonitis has established an association with mortality. [10] Sometimes severe pneumonitis may misguide surgeon in diagnosis, which may lead to delayed diagnosis. However, to reduce mortality among EF patients, respiratory complications should be prevented.

Out of all included patients, 5 (28%) patients need pre-operative ventilator care. Mortality among them was 80% (4 out of 5) but difference was found statistically non-significant. Overall mortality in the present study was 77.8% (14 out of 18). Mortality was high among patients with low birth weight, long gap length, presented with respiratory complication and high age in the presentation. No control group was the major limitation of this study. Comparison of this group was done with a group of patients who presented earlier.

CONCLUSION:

To conclude, considering high mortality among EA patients with late presentations in EA some pre and post-operative factors can be taken under concern. High mortality was observed in patients with associated congenital anomalies, wide gap length, low birth weight, respiratory complications at the time of presentation, Waterston C classification, pre-operative ventilator care.

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