Original Article

Early Outcome of Surgical Intervention of Esophageal Atresia and Tracheo-esophageal Fistula in Erbil Pediatric Surgical Center

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Abstract

Background: Esophageal atresia (EA) and treacheo-esophageal fistula occur in 1 out of every 3500 live births. Children born with EA have a higher incidence of prematurity than the general population EA. The treatment of EA and tracheo-esophageal fistula, although still a challenge, represents one of the true successes of newborn surgery. **Objective:** The aim of this study cases with EA and/or tracheo-esophageal fistula in Rapareen pediatric surgery center, Erbil, Iraq, regarding management, and early outcome. **Materials and Methods:** Fifty-three neonates were enrolled in this study from October 2011 to September 2015. Preoperative investigations included chest X-ray, ultrasound of the abdomen, and echocardiography. All patients were resuscitated before surgical intervention. Statistical Package for the Social Sciences version 20 was used for data analysis. **Results:** Out of 53 cases, 21 survived and 32 died. Thirty patients were male and 23 were female (male-to-female ratio 1.3:1). Twelve were premature and 41 term babies. The most common type was EA and distal fistula in 47 cases, pure atresia in 5 cases, and EA with both distal and proximal fistula in one case. Presenting features were excessive salivation in all cases, failure to pass nasogastric tube in 98.1%, cyanosis in 69.8%, and chocking in 37.7%. Prenatal history of polyhydramnios was present in 67.9%. Eighteen cases had associated anomalies, most of them were cardiac. **Conclusion:** EA with distal tracheo-esophageal fistula is the most common type of anomaly. Early diagnosis, weight, maturity, and associated anomalies are the most important factors that affect the outcome. Postoperative respiratory care is necessary, especially for those who have a preoperative chest infection.

Keywords: Esophageal atresia, neonates, tracheo-esophageal fistula

Introduction

The treatment of esophageal atresia (EA) and tracheo-esophageal fistula, although still a challenge, represents one of the true successes of newborn surgery.[1] The development of EA/tracheo-esophageal fistula (EA and TEF) appears to be a complex multifactorial process involving genetic and environmental factors to some degree. Up to 10% of patients with EA have a defined genetic syndrome that can be diagnosed, leaving the remaining 90% with an unknown etiology for their malformation.[2] EA and TEF occurs in 1 out of every 3500 live births.[3] Children born with EA have a higher incidence of prematurity than the general population.[4] EA and TEF present in two forms, either in isolation (50%) or as a syndromic form (50%) associated with other abnormalities.[5]

EA and TEF present in many forms and various classification systems have been used to describe them. For clarity, it seems much better to give descriptive names to the major subtypes.[6]

Prenatal ultrasonography attempts to predict the presence of EA using the combination of polyhydramnios, absent or small stomach bubble, and an "upper pouch" sign.[7] Infants born with EA and TEF have difficulty handling their oral secretions and will choke, cough, and possibly become cyanotic with their first feeding.[8]

After a feeding attempt, they will spit-up undigested formula or breast milk. This leads to an attempt to place a tube in the stomach, which does not travel as far as expected and meets resistance. A radiograph of the chest and abdomen shows the tube coiled in the esophageal pouch in the upper mediastinum,

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Access this article online

Quick Response Code:

Website: www.medjbabylon.org

DOI: 10.4103/MJBL.MJBL_39_18

How to cite this article: Hamad NA, Kak-Ahmed HA, Allaquli NI. Early outcome of surgical intervention of esophageal atresia and tracheo-esophageal fistula in erbil pediatric surgical center. Med J Babylon 2018;15:129-34.

confirming EA. The diagnosis is made and the remainder of the preoperative evaluation attempts to define a proximal fistula between the esophageal pouch and the trachea and to characterize associated anomalies.[9] A tracheo-esophageal fistula without EA (H-type fistula) may not present in the initial neonatal period and can be more difficult to diagnose. A tube will go into the stomach if passed, but persistent coughing and choking with oral feeds should prompt a search for an isolated fistula. A prone pull-back esophagram or bronchoscopy with esophagoscopy using methylene blue as a dye is used to find an isolated fistula. Placement of a catheter across the fistula during bronchoscopy aids the dissection greatly.[10]

In healthy newborns, the repair can be done in the first 24 h of life to minimize the risk of aspiration and the resulting pneumonitis. The goal of surgical therapy is to divide and close the fistula between the trachea and esophagus, and return continuity to the esophagus. In most situations, a primary repair is feasible.[11]

Recently, thoracoscopic repair of EA and tracheo-esophageal fistula has proven feasible with similar outcomes to those seen with open repair.[12] After surgery (whether open or thoracoscopic) the infant returns to the Intensive Care Unit and continues on intravenous nutrition and antibiotics. Feedings may be started through a trans-anastomotic tube 2–3 days after surgery. On the postoperative days 5–7, esophagography can be used to check the integrity of the anastomosis. If no leak is seen, feedings are started orally, and the chest tube removed the following day.[13]

The outcome for infants with EA and TEF has improved over time to the point where, unless the infant has major cardiac anomalies, significant chromosomal abnormalities, severe pulmonary complications, or a birth weight <1500 g, he/she will survive. The long-term problems after EA and TEF repair include pulmonary issues (especially reactive airway disease, bronchitis, and pneumonias) and upper gastrointestinal complaints of dysphagia and gastroesophageal reflux.[14]

The aim of this study was to evaluate patients with EA and tracheo-esophageal fistula who underwent an operation in Erbil pediatric surgical center regarding clinical presentation, diagnosis, types, management, and early outcome.

Materials and Methods

This observational study was conducted in the department of pediatric surgery, Rapareen pediatric teaching Hospital/Erbil, Iraq, from October 2011 to September 2015. A total of 53 neonates were included in the study.

A special data collecting form has been used including name, age, sex, gestational age, birth weight, residency, the age of the mother, family history, prenatal history, type of the anomaly, clinical presentation, associated anomalies, operation and operative findings, postoperative management, complications, and outcome.

The diagnosis was based on history and clinical examination, aided by radiological examination through plain X-ray after inserting a nasogastric (NG) tube. All patients admitted, were managed and resuscitated by putting them in incubators with continuous oxygenation, intravenous fluids, antibiotics, suctioning of upper esophageal pouch and mouth, and Vitamin K administration.

Ultrasound of abdomen and echocardiography were used routinely to exclude associated cardiac and intraabdominal organ anomalies, especially renal anomalies.

Blood analysis was sent for Hb, BG, and Rh, TSB, RBS, serum electrolytes, urea, and creatinine for all of the cases.

Right thoracotomy was done for most of the cases with ligation of fistulous connection between the trachea and lower esophagus and primary anastomosis between proximal and distal ends of the esophagus, except for one case whose aortic arch was in the right, left thoracotomy was performed. Neonates with long gap proximal and distal ends as in most of the isolated type were managed by esophagectomy and gastrostomy.

Postoperatively, most of the cases were admitted to the respiratory care unit to be at rest with ventilatory support and anesthesia for at least 24 h. Almost all postoperative complications were minor which dealt with nonoperatively. Oral feeding started in the 3rd–5th day in most of the cases, patients who had leaks or who were kept NPO for more than 7 days were treated with total parenteral nutrition.

Data entry and analysis

Each returned questionnaire was given an identity number. Before data entry and analysis, the questions of the study were coded. The data were entered into a Microsoft Excel Spreadsheet, after data cleaning; the data were transported into Statistical Package for the Social Sciences version 20 (IBM, Armonk, NY, United states of America).

Descriptive statistics (frequencies and percentages) were calculated for all variables, as well as analytical statistics was done to find the relations between categorical variables using Chi-squared test, and Fisher's exact test. A value of $P \leq 0.05$ was considered as statistically significant.

Results

Thirty (56.6%) patients were males and 23 (43.4%); patients were females with male-to-female ratio 1.3:1.

Of 53 cases studied, the weight of 11 (20.8%) cases was < 2.5 kg, eight (15.2%) cases were 2.5 kg. Thirty-four (64.2%) cases were more than 2.5 kg. The mean weight was 2914 ± 572 g [Figure 1].

Regarding the distribution of the cases according to the maturity, 41 (77.4%) cases were full-term babies, whereas 12 (22.6%) cases were preterm babies (<37 weeks gestation).

Eleven (20.8%) cases presented in the 1st day of life, most of the cases were more than 48 h at the time of presentation. The mean age was 48 h, ranging from 6 h to 13 days [Table 1].

Excessive salivation was the presenting feature in all cases (100%), and failure to pass NG tube in all (98.1%), except one case (1.9%), which presented with pneumoperitoneum due to gastric perforation diagnosed at laparotomy (1.9%) [Table 2].

Polyhydramnios was present in 33 (62.3%) of mothers prenatally. One mother was taking antithyroid drug prenatally [Table 3].

Regarding the distribution of the cases according to the mode of delivery, 47% were delivered by cesarean section and 53% by normal vaginal delivery.

Maternal ages ranges from 21 to 30 years in (52.8%) of cases. Only two mothers were above 40 (3.8%) [Table 4].

Eighteen (34%) cases had associated anomalies. Cardiac anomalies were the most common anomaly (78%) among all system anomalies. Eleven neonates (20.8%) had more than one cardiac anomaly [Table 5].

Out of 53 patients, 47 cases had proximal atresia with distal fistula (Type C) [Table 6].

Regarding the distribution of the cases according to the survival and deaths, Among 53 recorded cases, only 21 (39.6%) cases lived, and 32 cases (60.4%) died postoperatively (death after discharge from hospital is not included).

As reported in literature outcome depends mainly on maturity, weight, and associated cardiac anomalies. In this study, we correlated these risk criteria with survival on our patients [Tables 7-9]. The cases of esophageal atresia/tracheo-esophageal fistula of the pediatric patients in the present study, are represented in Figures 2-5.

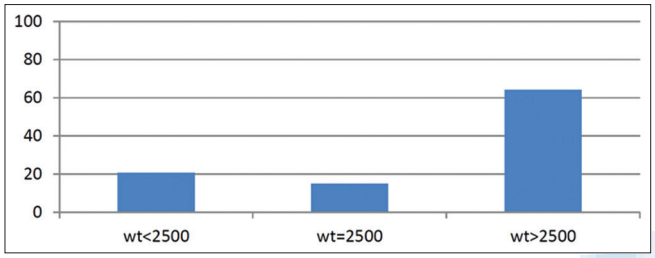

Figure 1: Distribution of the cases according to the body weight of babies in gram

Table 1: Distribution of the cases according to the age at presentation to surgical unit

Age at presentation*	*n* (%)
<24	11 (20.8)
24-48	17 (32.1)
>48	25 (47.2)
Total	53 (100)

*Age of the baby (h)

Table 2: Distribution of the cases according to the mode of presentation

Mode of presentation	Number of patients (%)
Excessive salivation	53 (100)
Failure to pass nasogastric tube	52 (98.1)
Chocking	37 (69.8)
Cyanosis	11 (20.8)
Pneumoperitoneum	1 (1.9)

Table 3: Distribution of the cases according to the prenatal history

Prenatal history	Number of patients (%)
Polyhydramnios	33 (62.3)
Maternal HTN	7 (13.2)
Drugs	8 (15.1)
Hyperthyroidism	1 (1.9)

HTN: Hypertension

Discussion

In this study, 30 (56.6%) neonates were male and 23 (43.4%) were female, with male: female ratio was 1.3:1, which is similar to C. Bode *et al*.[15] (whose cases were 25) in which 56% were male and 44% were female with male: female ratio of 1.2:1. and Tandon *et al*.[16] (127 patients), (66% of males and 34% of females).

In our set up, 64.2% of cases weighing > 2500 g, slightly more than that of Tandon *et al*.[16] study in which most of the neonates (53%) were below 2500 g. This is due to a limited number of cases in our study in comparison to their study.

Table 4: Distribution of the cases according to the maternal age

Age of the mother (years)	Number of mothers (%)
15-20	8 (15.1)
21-30	28 (52.8)
31-40	15 (28.3)
41-50	2 (3.8)
Total	53 (100)

Table 5: Distribution of various congenital anomalies

Types of congenital anomalies	Number of cases (%)
Cardiac anomalies	14 (78)
Imperforate anus	1 (5.5)
Down syndrome	1 (5.5)
Hydrocephalus + ambiguous genitalia	1 (5.5)
Solitary kidney + polydactyly	1 (5.5)
Total incidence	18 (100.0)

Table 6: Distribution of the cases according to the anatomical types

Types (gross)	Frequency (%)
A	5 (9.4)
C	47 (88.7)
D	1 (1.9)
Total	53 (100.0)

Table 7: Relation between associated congenital anomalies and survival

Associated congenital anomalies	Survival		Total (%)	*P*
	No (%)	Yes (%)		
No	19 (35.8)	16 (30.2)	35 (66)	0.2
Yes	13 (24.6)	5 (9.4)	18 (34)	
Total	32 (60.4)	21 (39.6)	53 (100)	

Table 8: Relation between maturity and survival

Maturity	Survival		Total (%)	*P*
	No (%)	Yes (%)		
Preterm	11 (20.8)	1 (1.9)	12 (22.6)	0.01
Term	21 (39.6)	20 (37.7)	41 (77.4)	
Total	32 (60.4)	21 (39.6)	53 (100)	

Table 9: Relation between weight and survival

Weight	Survival		Total (%)	*P*
	No (%)	Yes (%)		
<2500	8 (15.1)	3 (5.7)	11 (20.8)	0.7
2500	5 (9.4)	3 (5.7)	8 (15.1)	
>2500	19 (35.8)	15 (28.3)	34 (64.1)	
Total	32 (60.4)	21 (39.6)	53 (100)	

Full-term babies constituted 77.4% of our patients and 22.6% were premature which is comparable with the study done by Bode *et al*.[15] 40 (15 cases) and Tandon *et* al.[16] in which 80%–69% of cases were full-term, respectively.

In this study, most of the patients (47.2%) were diagnosed or suspected to have EA and TEF after 48 h after delivery, only 20.8% were diagnosed in the 1st day. This is slightly different from that of Tandon *et al*.[16] in which 36% diagnosed in the 1st day of life, this may be due to poor prenatal assessment, postnatal evaluation by a pediatrician, neonatal care, or parental education.

Excessive salivation was the presenting feature in all cases (100%). Moreover, failure to pass NG tube in all except one case (98.1%), which became standard diagnostic criteria, chocking with feeding was present in 69.8% of cases which is the same to Bianca and Ettore study[17] (90 patients collected), the diagnosis was suspected after choking with feeding and usually confirmed by the inability to pass NGT into the stomach.

Polyhydramnios was positive in 33 (62.3%) cases, this is much more than of Verma *et al*.[18] (50 cases) and Anwar-Ul-Haq *et al*.[19] (80 cases) studies (10% and 16%, respectively), but more similar to that reported in the literature.

The mothers of 8 cases 15.1% gave a history of drug ingestion. Drugs in 7 patients were antihypertensive and in 1 patient antithyroid drug, but we do not know the relation between those drugs and this anomaly and we cannot trace any paper related to drug as a predisposing factor.

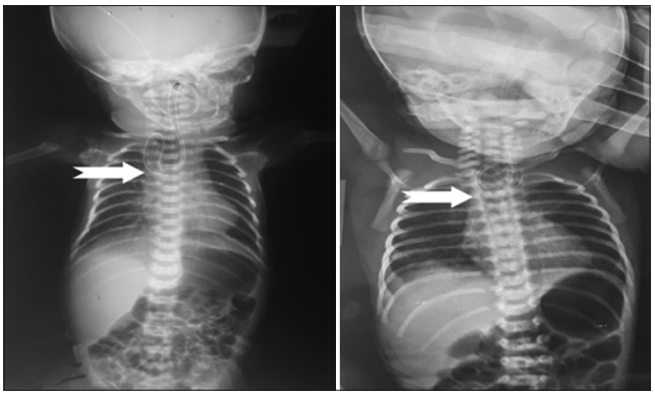

Figure 2: Chest X-ray in two different cases of esophageal atresia/tracheo-esophageal fistula shows coiled nasogastric tube (arrows) in upper pouch of esophagus take preoperatively from Rapareen pediatric teaching hospital

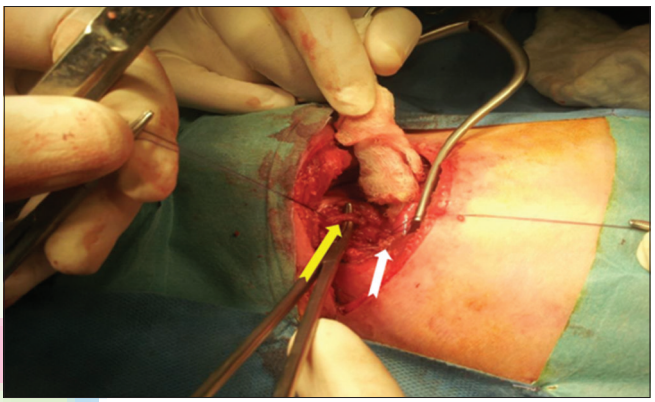

Figure 3: A patient with common type esophageal atresia/tracheo-esophageal fistula, Right thoracotomy extrapleural approach, see proximal pouch (white arrow), and distal fistula (yellow arrow). From Rapareen pediatric teaching hospital

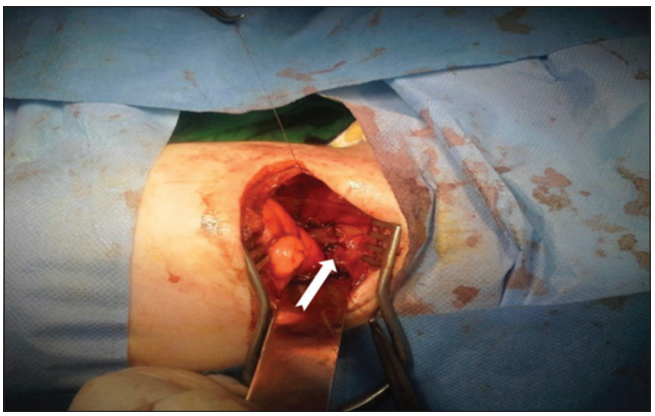

Figure 4: Primary end-end anastomosis (arrow). From Rapareen teaching hospital

Maternal age is not significant statistically for all groups, in our study the average maternal ages were 27.7 ± 6.5 years which is similar to Bianca and Ettore study[17] (28 ± 5.8 years).

In this study, 18 (34%) neonates had associated congenital anomalies which are compatible with Tandon *et al*.[16] Among

all anomalies, cardiac anomalies constituted 78% close to Yang *et al*.[20] (53.3%), but much more higher than Singh *et al*.[21] (301 patients) and Tandon *et al*.,[16] this may be due to some type of anomalies that are not included in their studies like PDA.

EA with tracheo-esophagel fistula (type C according to Gross classification) is the most common type in all literature and studies.[16] In this study, 47 (88.7%) cases had a common type (Type C), pure type (Type A) in 5 cases (9.4%), and both end fistula (Type D) in one case (1.9%). These results are compatible with most studies, but we had no tracheo-esophageal fistula without atresia (Type E), this may be because of a limited number of patients collected or not diagnosed and treated with upper respiratory tract infection.

In this study, mortality was high 60.4%, this is close to that of Anwar-Ul-Haq *et al*.[19] The causes of death in our study were chest infection, sepsis, and associated major cardiac anomalies [Table 10].

In our center, we used right thoracotomy, extrapleural approach and primary anastomosis by 4-0/5-0 vicryl as standard operative management of EA and TEF. All (100%) cases in our set up underwent surgery. Fifty (94.3%) patients operation performed through the right thoracotomy. In two (3.8%) patients, left thoracotomy performed because of the right aortic arch. One (1.9%) neonate presented with peumoperitonium, laparotomy done for him, gastric perforation found with air leak through the perforation site with assisted inflation, and hence, EA and TEF diagnosed intraoperative, gastrostomy done with esophagostomy.

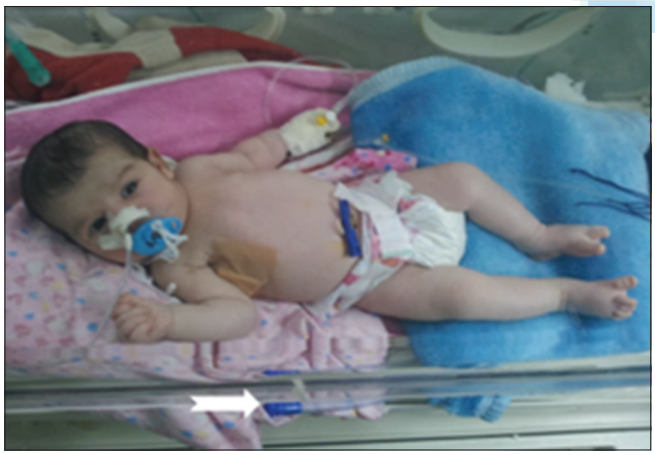

Figure 5: A postoperative baby in incubator with oxygen, nasogastric tube and chest tube (arrow)

Table 10: Comparing our outcome with other studies

	Present study (*n*=53), *n* (%)	Anwar-Ul-Haq *et al*. (*n*=80), *n* (%)	Verma *et al*. (*n*=50), *n* (%)	JH Seo *et al*.[23] (*n*=97), *n* (%)
Deaths	32 (60.4)	47 (59)	20 (40)	20 (20.6)

Out of other 52 cases, eight (15.4%) cases had long gap distal and proximal segments of the esophagus (4 patients were of pure type), and hence staged repairs performed. Six of them gastrostomy and esophagostomy done, the other two neonates traction sutures (Foker's procedure) applied to a proximal segment of the esophagus with gastrostomy.

Other 44 (84.6%) patients had a short gap, and hence, primary anastomosis performed, with NG tube bypassed across the anastomosis, chest tube left for some time of hospital stay.

These results were close to the study of Tsai *et al*.,[22] in which out of 68 patients, 44 (67.4%) patients underwent primary esophageal anastomoses, 7 (10.3%) patients underwent delayed primary anastomoses, and 5 (7.4%) patients underwent repair of H-type fistulas. Twelve (17.6%) patients underwent staged repairs for long-gap type A and C anomalies. Primary repair included fistula division and end–end esophagoesophageal anastomoses within the first 48 h. Delayed primary repairs included fistula division and esophageal anastomoses after 48 h after an initial gastrostomy.

Staged repairs include initial thoracotomy and fistula division with gastrostomy or esophagostomy, and delayed esophageal anastomosis. Primary repairs were performed by a retropleural approach whenever feasible. A single layer end–end esophageal anastomosis was constructed using interrupted 5-0 silk sutures.

The difference with his study, in the present study, is that delayed primary anastomosis is not attempted, and we had no H-type TEF cases, this is may be due to a limited number of patients in our study. Furthermore, we have no experience with silk material for anastomosis instead we used vicryl. The results of this study were also compatible with those of Seo *et al*.[23] who studied the tracheoesophageal fistula and esophageal atresia in Korean pediatric patients.

Conclusions

EA with distal fistula was the most common type in this study. Excess Frothy secretion from the mouth with failure to pass NG tube was the main presenting features. The radiological study (Plain) is an important diagnostic tool through which one can diagnose such anomaly, some of the associated anomalies and knowing the type of EA while contrast study is necessary in equivocal cases. The high mortality rate (60.4%) in this study needs further evaluation of the causes. Primary anastomosis is used in all patients of a common type of EA in our center.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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