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Surgical Etiology of Respiratory Distress in Infant and Short Term Outcome at Benghazi Children Hospital (2017-2018)

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Abstract **Original Research Article**

Background: Respiratory distress (RD) is a common problem in newborns immediately following birth. It is often seen during the transition from fetal to neonatal life. ***Aims:*** This study was conducted to demonstrate the surgical causes of respiratory distress in infants admitted to paediatric surgical department of Benghazi Children Hospital and to describe the short term outcome of the sample. ***Methodology:*** A retrospective observational descriptive cross section study was done to achieve the aims of this study. ***Results:*** A sample of (50) children admitted with respiratory distress due to congenital anomalies, the age range of the sample was from one day to one year, male were (70%) and females and (30%). Residency (20%) from Benghazi and (80%) from outside Benghazi. Normal delivery was reported in52% of the cases and (48%) C/S, (86%) were born at term and (14%) preterm. The mean weight of the cases was (3.38) (±1.91) kg with minimum of (2) kg and maximum (10) kg. TOF in (52%), diaphragmatic hernia in (40%) and lung emphysema in (8%). Clinical presentation of the cases was mainly with respiratory distress in (84%), while (16%) presented with frothy secretion, ultrasound scan was done to (88%) and was normal. Echocardiogram was normal, atrial septal defect in (16%), ventricular septal defect in (14%), patent ductus arteriosus in (8%). (80%) were operated. Death was recorded in half of the cases. Cases with TOF were in total (26) cases of them (17) died and (9) survived, diaphragmatic hernia cases were (20), of them (12) discharged and (8) died, all cases of congenital lobar emphysema discharged. This association was highly significant Fisher`s (exact=6.9 P=0.005). all preterm neonate died (seven cases) and those who were term were in total (43) of them (25) survived and (18) died, Fisher`s (exact=10.85 P=0.005). Cases who were presented with respiratory distress were in total (42) of them (18) died and (25) discharged, while cases who presented with frothy secretions were in total (8) of them (7) died. Fisher`s (exact=5.4 P=0.022). cases with normal echo were (25) cases of them (19) discharged and only (6) died, cases with ASD were (8) of them 6 died, (7) cases with VSD all of them died, PDA associated with good outcome as they were four cases all of them discharged, and echo was not done to (6) cases and all of them had died Fisher`s (exact=25.6 P=0.001). Operation for congenital anomalies was done to (40) cases of them (16) died and (24) discharged, and not done to (10) cases of them (9) died and one case discharged. This association was with highly statistical significance. Fisher`s (exact=8, P=0.005). ***Conclusion:*** RD in neonate was more common in male than female; cases from outside Benghazi were more frequent. Predicting cases outcome was not based on variables such as gender, mode of delivery, city of residence which showed no significant association with cases outcome. Poor outcome predictors of the current study were diagnosis, birth maturity, clinical presentation, ECHO results.

Keywords: Respiratory distress, tracheoesophageal fistula, atresia, diaphragmatic hernia.

INTRODUCTION

Respiratory distress (RD) is a common problem in newborns immediately following birth. It is often seen during the transition from fetal to neonatal life. RD may be transient in some newborns, if it persists, then there is a need for proper diagnostic and therapeutic interventions to optimize outcomes and minimize morbidity [1]. RD is one of the most common reasons for neonates to be admitted to the neonatal intensive care unit (NICU) [2, 3]. It was estimated that (15%) of term infants and (29%) of late preterm infants

admitted to the NICU develop significant respiratory morbidity. This incidence is even higher for infants born before (34) weeks' gestation [4, 5].

Certain risk factors increase the likelihood of neonatal RD. these risk factors include; prematurity, low Apgar scores, meconium aspiration syndrome, caesarian section delivery, gestational diabetes, maternal chorioamnionitis, premature rupture of membranes [6], and oligohydramnios, as well as structural lung abnormalities are some predictors identified in previous studies [7-10]. Other common causes include transient tachypnea of the newborn, pneumonia, sepsis, pneumothorax, and persistent pulmonary hypertension of the newborn [11]. Regarding surgical causes of respiratory distress in infants the underlying mechanisms include airway obstruction, pulmonary collapse or displacement and parenchyma disease or insufficiency; the common causes are congenital diaphragmatic hernia, congenital cystic adenomatoid malformation, congenital lobar emphysema and esophageal atresia with or without trachea -esophageal fistula. Obstruction lesion of the newborn airway include choanal atresia, macroglossia, Pierre - Robin syndrome, lymphangioma, teratoma or other mediastinal masses, cysts, subglottic stenosis and laryngo Tracheomalacia [2, 3].

These surgical causes, if not recognized and managed quickly, RD can escalate to respiratory failure, cardiopulmonary arrest, and even death. Therefore, it is imperative that any health care practitioner caring for newborn infants be able to readily recognize the signs and symptoms of RD, differentiate the various causes, and initiate management strategies to prevent significant complications or death [12, 13].

Globally, there are different policies, strategies, and programs which work on or advocate for the prevention and care of preterm neonates and their birth outcomes, including RD, like the Sustainable Development Goals (SDGs) and the Every Women and Every Child initiative [14, 15]. Despite these efforts, RD remains among the leading causes of neonatal mortality and morbidity [16, 20]. Indeed, in Ethiopia, RD is the most common cause of neonatal mortality and morbidity [21] resulting in exponentially increasing neonatal care costs within the first 28 days of life. Additionally, few studies have been conducted in developing countries to assess RD in these regions, including Ethiopia. Therefore, this study we aimed to determine the incidence and predictors of RD among neonates who were admitted to the NICU at Benghazi Children Hospital [21].

REVIEW OF LITERATURE

Respiratory distress in term infants is still a significant cause of admission to NICU and a predisposing factor for neonatal mortality and morbidity. Preventative and anticipatory measures should be further explored to decrease the burden of this health problem [22].

Surgical congenital causes of neonatal RD sometimes complicated and difficult to diagnose, laryngeal atresia (LA) and tracheal agenesis (TA) are two of the rare causes of intrinsic airway obstruction congenital high airway obstruction syndrome (CHAOS) is a rare and frequently fatal disorder caused by complete or near - complete obstruction of the fetal upper airway [23]. CHAOS often results in stillbirths, while severe respiratory distress, hypoxia, absence of audible cry, and failure to intubate the airway are typical clinical features in newborn survivors, immediately after delivery. Therefore, prenatal diagnosis is desirable, even though difficult, in order to plan for ex - utero intrapartum treatment [24, 25]. LA seldom allows for fetal intervention and tracheostomy at birth, TA is almost universally fatal even in highly specialized centers, and survivors are exceptional. In a systematic review, Smith *et al.*, reported a mortality rate of (87.2%) within the first week of life and (92.6%) at 1 year; (41.3%) of patients did not survive beyond the first (24) h of life [26].

TA is a rare congenital airway malformation, characterized by complete or partial absence of the trachea, frequently associated with carino - esophageal fistula, bronchoesophageal fistula, or tracheo - esophageal fistula (TEF). TA was first described by Payne in (1900) and later classified into three types by Floyd.

The typical presentation of TA started immediately after delivery, the newborn becomes cyanotic and showed severe respiratory distress. Bag - valve -mask ventilation did not relieve the respiratory distress but allowed for temporary oxygenation during subsequent unsuccessful oral tracheal intubation [27].

LA is usually sporadic, although it can manifest as part of syndromes or chromosomal anomalies, it is characterized by complete or near - complete laryngeal obstruction and is classified according to either the anatomic site of obstruction or the presence of an associated TEF and the following clinical presentations [24].

In year 1965 Smith & Bain classified LA as the following (smith, 1965 in Bresciani, 2021) [29].

Tracheoesophageal Fistula (TOF)

TOF/EA is a common congenital anomaly of the respiratory tract, with an incidence of 1 in (3,500) to 1 in (4,500) live births [30]. Though it is typically associated to other congenital abnormalities like cardiac defect, esophageal atresia (EA) and more complex syndromes, the isolated TOF accounts for (4%) of cases [31]. The defective lateral separation of the foregut into the esophagus and trachea during the embryonic period is the most commonly accepted pathogenic hypothesis,

that is also considered to explain the etiology of other airways or digestive malformations (picture 3) (laryngo - tracheoesophageal cleft, tracheal atresia or isolated esophageal stenosis).About half of all children born with TOF/EA have other health problems. This is called the VACTERL association. Each letter stands for a problem that may occur with TEF/EA: vertebral, problems with the spine, anal atresia, a problem with the way a baby's anus or rectum has formed, cardiac problems, tracheoesophageal fistula, renal, or kidney problems, limb, problems with arms and legs [32]. The clinical presentation of TEF varies according to the presence or absence of EA. Infants with TEF and EA usually become symptomatic immediately after birth, with drooling, choking and respiratory distress during swallowing. In addition, the air passing through the fistula leads to gastric distension and subsequent reflux of gastric contents through the same TOF, resulting in aspiration pneumonia.

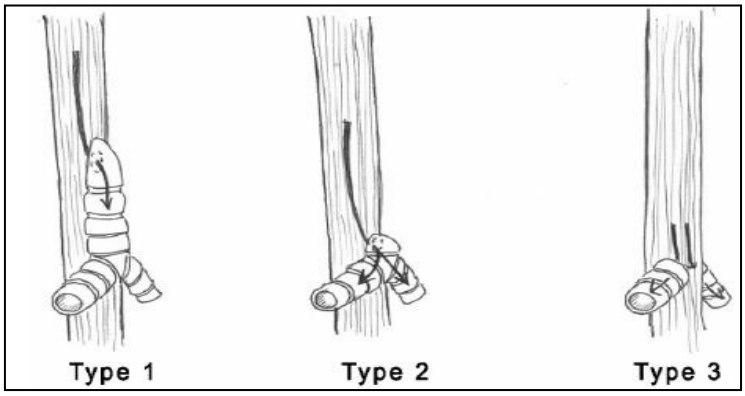

Figure 1: Floyd classification of TA (Bresciani, 2021: 2)

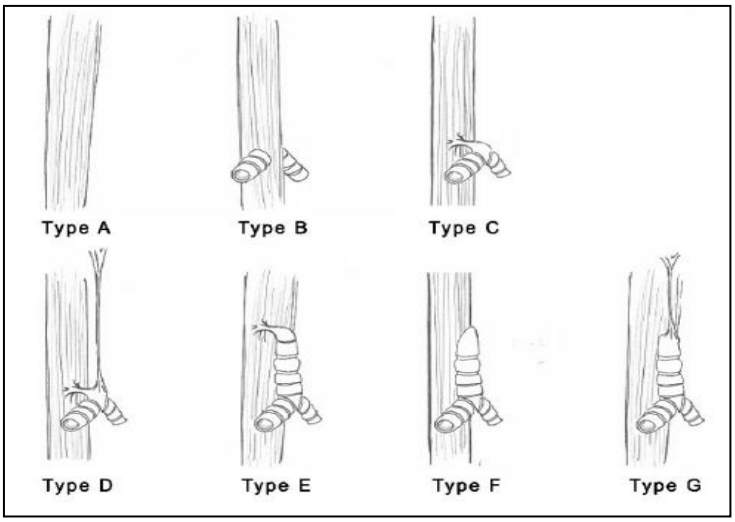

Figure 2: Faro classification of TA (Bresciani, 2021: 2)

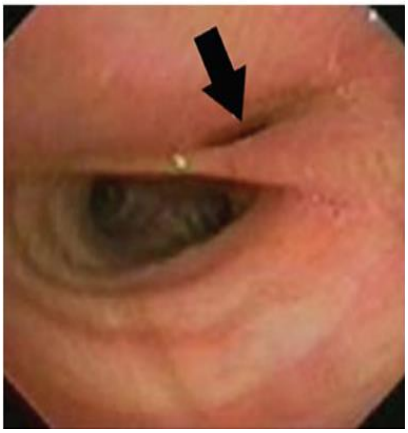

Figure 3: Trechio esophageal fistula

Classification of Esophageal Atresia (figure 4)

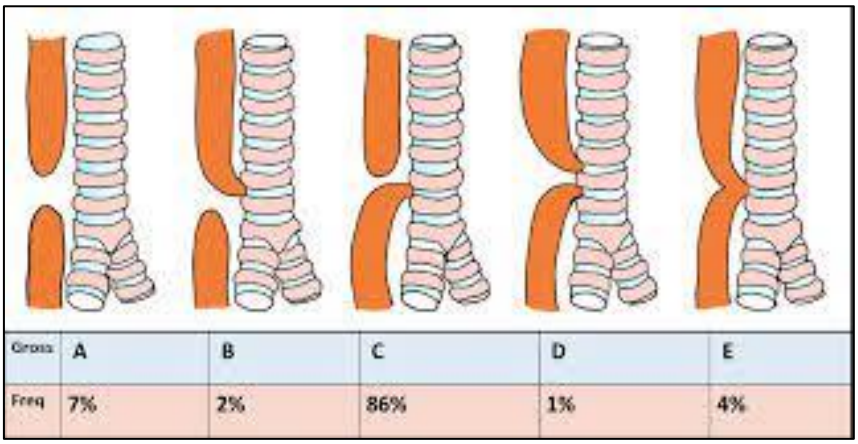

Figure 4: Types of oesophageal atresia

There are many forms EA and TOF can present in and different classification have been proposed and applied to describe them. Those disorders can be considered as anomalies spectrum (picture 4-1) [33].

Different classification system has been described by different authors with confusing definition including subclasses within major subtypes [34, 35].

Oesophageal Atresia with Distal Fistula (the Gross Type C):

This is the most common variety of EA-TOF, representing about85% of cases of EA [36]. The proximal esophagus becomes very dilated and its wall get thickened and descends into the superior mediastinum usually up to the level of the third or fourth dorsal vertebrae. The distal esophagus is slender with a thin wall. It opens posteriorly into the trachea either at the carina level or little higher. The length of esophagus varies from very short to quite tall .In very rare cases, the distal fistula may be occluded, which causes, the misdiagnosis of EA without distal fistula [41].

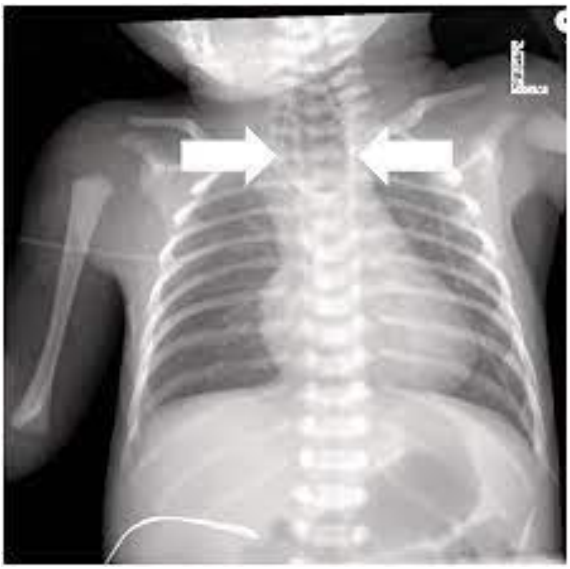

Figure 5: Plain x -ray of newborn with oesophageal atresia with distal fistula

Pure Esophgeal Atresia without fistula (The Gross Type A)

The incidence of this type rounds about (7%) both the proximal as well as distal esophageal ends blindly in the posterior mediastinum.

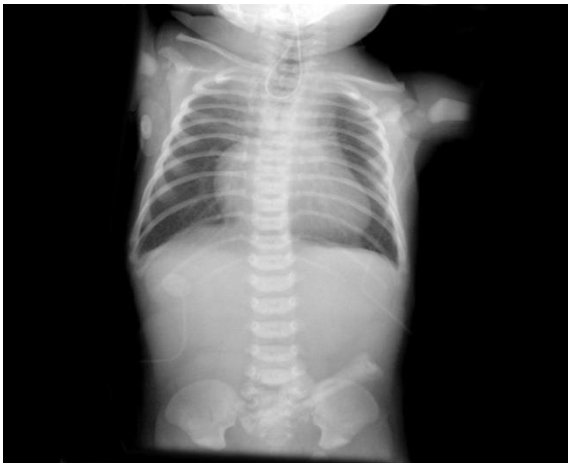

Figure 6: Plain x - ray of newborn with pure Oesophageal Atresia without fistula

H - Type fistula without Esophageal Atresia (the Gross Type E)

H -type TOF without atresia may be a component of the VACTERL association, so it is usually discussed in context of EA. The incidence of this type is about (4%). The fistula communicates the membranous trachea with the esophagus [37].

Esophageal Atresia with Proximal Fistula (the Gross Type B):

The association of a pure EA with proximal fistula is generally to be about (2%), but two and three have been also described. This fistula is similar to the H - type as it starts proximally on the trachea and ends distally in the distally in the dilated proximal esophagus [38].

Esophageal Atresia with Proximal and Distal Fistulas (the Gross Type D)

This variety is very uncommon one with an incidence of less than (1%). Also cases of EA with two proximal fistulas and distal fistula have also been reported [39, 40].

Diaphragmatic Hernia (CDH)

CDH is a developmental occurs when the diaphragm, the muscle that separates the chest from the abdomen, fails to close during prenatal development, commonly located on the left side, this defect allows passage of the abdominal viscera into the thorax. Leading to contralateral displacement of the mediastinum and hypoplasia of the lungs with abnormal arterioles causing pulmonary hypertension (Picture 4). As a result, respiratory and cardiovascular functions will be severely compromised at birth and this, together with the frequently associated malformations, cause considerable mortality and morbidity [41, 42].

Neonate with sever CDH will show very low APGAR scores, the symptoms of CDH are typical for respiratory distress with insufficient oxygenation, excavated abdomen with sternal protrusion and displacement of the heart sounds to the contralateral side [43, 44].

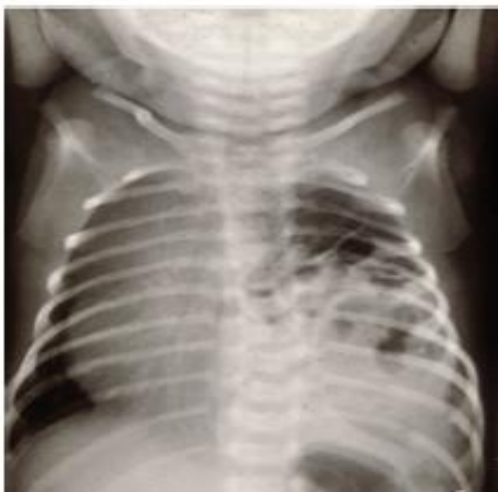

Figure 7: Plain X - ray of the thorax of a newborn with CDH. There are bowel loops into the left hemi - thorax, the mediastinum is displaced

Congenital Lobar Emphysema

Congenital lobar emphysema (CLE) is one of these rare and serious developmental lung malformations, occurring in (1:20,000) to (1:30,000) deliveries, and characterized by partial obstruction of the bronchus resulting in hyperinflation of lung lobe [45].

It is usually unilateral with a male pre ponderance. The most commonly involved lobes are the left upper lobe (40 - 50%) and the right middle lobe (30 - 40%) [46]. However, involvement of pulmonary segments [47], and bilateral lung involvement were reported [48]. CLE commonly presented at the neonatal period, but rare cases were reported in late childhood or early adulthood [49]. There is no uniform etiology of CLE. Up to (50%) of cases have no cause, while congenital defect of cartilage presents in (25%) and other causes of bronchial obstruction underlying the remaining (25%) of cases [50]. There is an additional evidence for inherited factors in the etiology of congenital lobar emphysema. Roberts *et al.*, [51] described two cases of CLE involving the right upper and middle lobes in a father and son secondary to relative deficiency of the bronchial cartilage. CLE may associate with other congenital cardiac malformations, such as patent ductus arteriosus, atrial septal defect, ventricular septal defect, total anomalous pulmonary venous return, and Tetralogy of Fallot [52].

In addition, CLE may associate with double superior vena cava, horseshoe kidney, and polysplenia [53, 54].

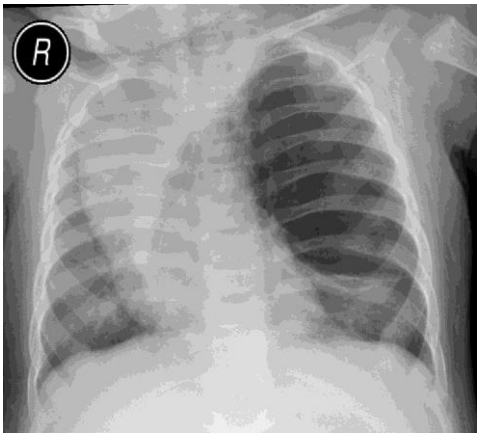

Figure 8: Infant with congenital lobar emphysema

Presentation and Diagnosis Lesions may be asymptomatic or present with respiratory distress in the newborn period [48]. Later, infants may experience dyspnea (57%) or recurring respiratory infection (28%). On CXR, hyperlucency of the affected lobes is the characteristic feature. The diagnosis may be confirmed on chest CT. The differential diagnosis includes type (1) unicystic congenital pulmonary airway malformation CPAM, pneumothorax, Swyer -James syndrome, bronchogenic cyst, and diaphragmatic hernia [55].

AIMS OF THE STUDY

This study was conducted to:

1. Demonstrate the surgical causes of respiratory distress in neonate and infant admitted to paediatric surgical department of Benghazi Children Hospital.
2. Describe the short term outcome of the sample.

PATIENTS AND METHODS

Study Design

A retrospective observational descriptive cross section study was done to achieve the aims of this study.

Settings

The study was carried out in Paediatric Surgery Department of Benghazi Children teaching hospital the period extending from 1st of January 2017 to end of last 2018.

Sample

The sample of the study consisted of all infant and neonate admitted to paediatric surgery department or as referred from outside the hospital during the study period. Sampling techniques was none random criterion based sample.

Exclusion Criteria

1. The patient with medical causes of RD.
2. Files with incomplete data.

Data Collection

The data was collected by retrieving the medical records of the patients by using a well-designed Performa which included questions about child`s age, Age at admission, gender, residence of family, date of admission and date of discharge or death.

Neonatal data including birth weight, gestational age, mode of delivery vaginal or caesarean section.

Data of medical examination of the sample were recorded in detail including vital signs, dysmorphic feature, body weight, cardiovascular and respiratory system examination. The cause of distress was identified depending on the clinical criteria, laboratory workup and imaging studies.

Their initial relevant investigation included complete blood picture, blood glucose level, renal function tests, chest x ray, Abdominal ultrasound, MRI, and CT if performed according to the presentation to confirm the underlying causes of RD.

Data Analysis

The collected data were entered to statistical package for social sciences software for windows (SSPS) version (25), statistical analysis and calculation was performed as appropriate to describe the sample and summarize their features in term of figures and table as needed, finding any significant associations between cases outcome and clinical features was done using Chi square test and Fisher Exact test for categorical variables, significance level of (<0.05) is the cut off for judging relevance.

Ethical Considerations

The records of the sample were accessed after getting an official permission from authorized personals of statistics and archive department of Benghazi Children Hospital.

Patients names and entities were secured, data was analysed anonymously.

Descriptive Results

This study included a sample of (50) children admitted with respiratory distress due to congenital anomalies, the age range of the sample was from one day to three years (table 1, figure 9), (44%) of the sample (22/50) were just born and their age was one day, two cases aged two days and another two aged three days. Four cases were four days, one case five days, two cases were seven days, one case for each of (8) days, (15) days, (40) days, (45) days (2) months and nine months, two cases were seven months and another two eight months, six cases were one year, one case was three years of age (table 1, figure 9).

Gender of the sample study was predominantly male as (70%) of them were males (35/50) and (30%) (15/50) were females (table 2, figure 10).

Residency of the sample was either from Benghazi in (20%), (10/50) or from outside Benghazi in (80%) (40/50) cases (table 2).

Normal delivery was reported in 52% of the sample (26/50), and C/S in (48%) (24/50). (86%) of the sample delivered at term (43/50) compared to (14%) (7/50) were preterm (table 3).

The mean weight of the cases was (3.38) (±1.91) kg with minimum of 2 kg and maximum (10) kg (table 4).

Diagnosis of the cases of this study was as the following, TEF in (52%) of the cases (26/50), diaphragmatic hernia in (40%) (20/50) and lung emphysema in (8%) (four cases) (table 5, figure 11).Clinical presentation of the cases was mainly with respiratory distress which occurred in (84%) of the sample (42/50), while (16%) (8/50) presented with frothy secretion (table 5).

Radiological investigations were reported in the study sample (table 6), starting by abdominal ultrasound scan, which was done to (88%) of the sample (44 cases) and revealed normal results and not done to (12%) (6 cases). Echocardiogram was normal in (50%) of the cases (25 cases), revealed atrial septal defect in (16%) of the sample (8 cases), ventricular septal defect in (14%) of the sample (7 cases), patent ductus arteriosus in (8%) of the sample (4 cases) and not done to (12%) of the sample (6 cases) (table 6).

Most of the cases were operated for the congenital anomalies as they represented (80%) of the sample (40 cases) whereas the remaining (20%) (10 cases) were not operated (table7). Death was recorded in half of the cases (25/50) and the other half was discharged (table 7).

Inferential Results

Study of associations between outcome and determinant factors of this study showed that:

Diagnosis of The cases were an important determinant of The cases outcome, cases with TOF were in total (26) cases of them (17) died and (9) survived, diaphragmatic hernia cases were (20), of them (12) discharged and eight died, all cases of lung emphysema discharged. This association was highly significant Fisher`s (exact=6.9 P=0.005) (table 8, figure 12).

Birth maturity was another predictor of poor outcome as those who were preterm were all died (seven cases) and those who were term were in total (43) of them (25) survived and (18) died, Fisher`s (exact=10.85 P=0.005) (table8, figure 13).

Ultra sound scan was normal in (44) cases of them (19) died and (25) survived and discharged, while it was not done to six cases and all of died which could be due to inability to do it or immediate death of the child. Fisher`s (exact=6.8 P=0.011) (table 8, figure 14).

Cases who were presented with respiratory distress were in total (42) of them (18) died and (25) discharged, while cases who presented with frothy secretions were in total (8) of them (7) died. Fisher`s (exact=5.4 P=0.022) (table 8, figure 15).

Echocardiogram was another predictor of cases outcome, those who showed normal echo were (25) cases of them (19) discharged and only 6 died, cases with ASD were (8) of them 6 died, (7) cases with VSD all of them died, PDA associated with good outcome as they were four cases all of them discharged, and echo was not done to (6) cases and all of them had died Fisher`s (exact=25.6 P=0.001) (table 8, figure 16).Operation for congenital anomalies was done to (40) cases of them (16) died and (24) discharged, and not done to (10) cases of them (9) died and one case discharged. This association was with highly statistical significance. Fisher`s (exact=8, P=0.005) (table 8, figure 17).

Other variables such as gender, mode of delivery, city of residence did not show significant association with cases outcome.

Table 1: Age distribution and features of the study sample

Age	No.	%	Age statistics
1 d*	22	44.0	Mean age= 86.5 d(±159.2)
2 d	2	4.0	Median= 3 d
3 d	2	4.0	Mode = 1 d
4 d	4	8.0	Minimum =1 day
5 d	1	2.0	Maximum = 1 years
7 d	2	4.0	
8 d	1	2.0	
15 d	1	2.0	
40 d	1	2.0	
45 d	1	2.0	
2 m**	1	2.0	
7 m	2	4.0	
8 m	2	4.0	
9 m	1	2.0	
10m†	1	2.0	
1 y	6	12.0	
Total	50	100.0	

*= day, **= month, †= year

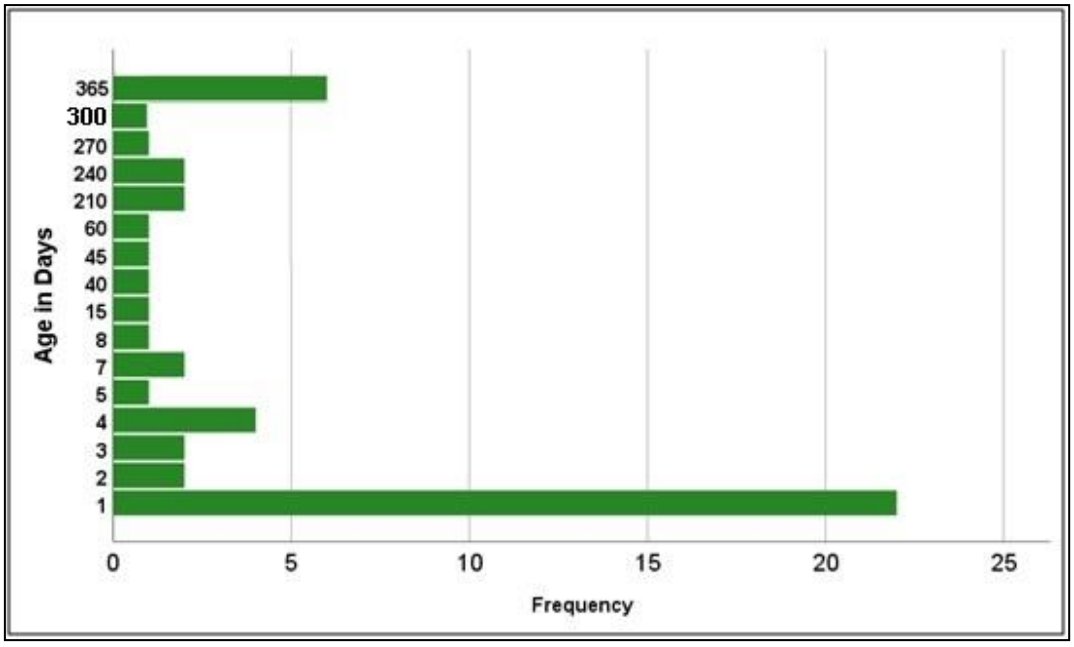

Figure 9: Age distribution of the study sample

Table 2: Gender and residence of the study sample

Characteristic		No.	%
Gender	Male	35	70.0
	Female	15	30.0
Residences	Benghazi	10	20.0
	Outside Benghazi	40	80.0

Table 3: Early life data of the study sample

Early life data		No.	%
Mode of delivery	Normal	26	52.0
	C/S	24	48.0
Birth Maturity	Term	43	86.0
	Preterm	7	14.0

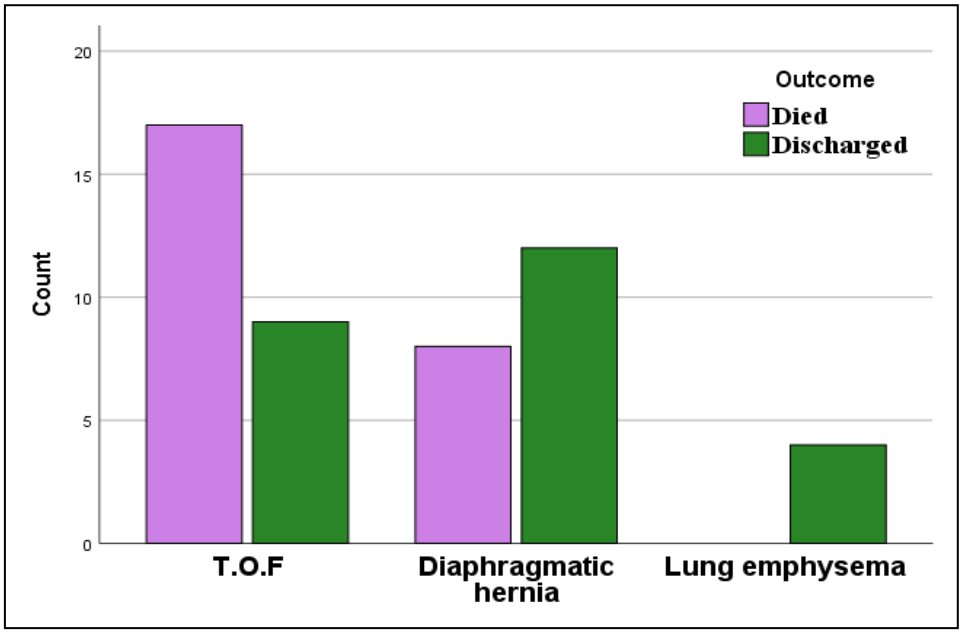

Figure 10: Diagnosis and outcome of the study sample

Table 4: Weight statistics of the study sample

Statistic	Value
N	50
Mean	3.38
Std. Deviation	1.91
Std. Error of Mean	0.27
Median	2.80
Mode	2
Minimum	2
Maximum	10

Table 5: Diagnosis and presentation of the study sample

Clinical characteristics		No.	%
Diagnosis	T.O.F	26	52.0
	Diaphragmatic hernia	20	40.0
	Lung emphysema	4	8.0
Presentation	R.D	42	84.0
	Frothy secretion	8	16.0

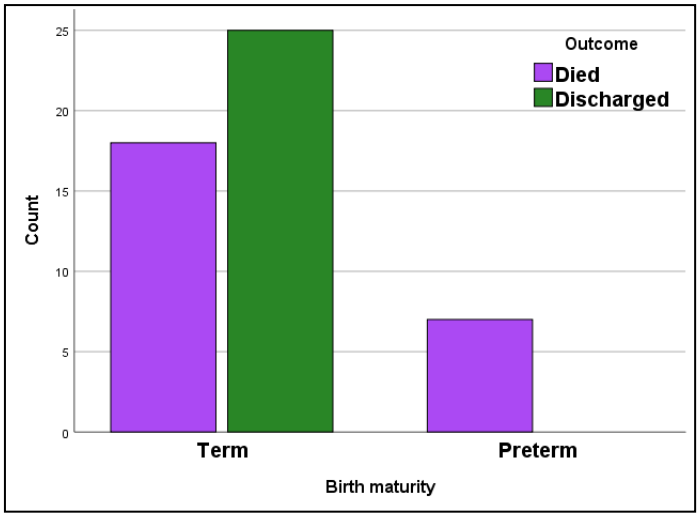

Figure 3: Birth maturity and outcome of the study sample

Table 6: Radiological investigations of the study sample

Radiological investigations		No.	%
USS Abdomen	Normal	44	88.0
	Not done	6	12.0
	Total	50	100.0
Echo	Normal	25	50.0
	ASD	8	16.0
	VSD	7	14.0
	PDA	4	8.0
	Not done	6	12.0

Table 7: Management and outcome of the study sample

Management and outcome		No.	%
Operation	Operated	40	80.0
	Not operated	10	20.0
End of management	Died	25	50.0
	Discharged	25	50.0

Table 8: The important determinants of outcome among the study sample

Clinical factor		Outcome		Total	Significance of difference
		Died (n=25)	Discharged (n=25)		
Diagnosis	T.O.F	17	9	26	Fisher`s exact=6.9 P=0.005**
	Diaphragmatic hernia	8	12	20	
	Lung emphysema	0	4	4	
Birth Maturity	Term	18	25	43	Fisher`s exact=10.85 P=0.005**
	Preterm	7	0	7	
USS Abdomen	Normal	19	25	44	Fisher`s exact=6.8 P=0.011*
	Not done	6	0	6	
Presentation	R.D	18	24	42	Fisher`s exact=5.4 P=0.022*
	Frothy secretion	7	1	8	
Echo	Normal	6	19	25	Fisher`s exact=25.6 P=0.001**
	ASD	6	2	8	
	VSD	7	0	7	
	PDA	0	4	4	
	Not done	6	0	6	
Operation	Operated	16	24	40	Fisher`s exact=8 P=0.005**
	Not operated	9	1	10	

* Significance result at P <0.05, ** Significance result at P <0.01

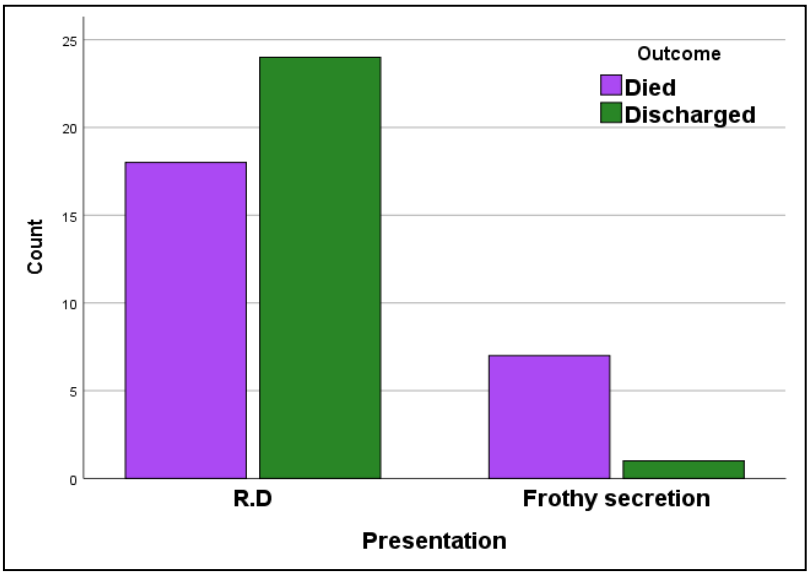

Figure 12: Presentations and outcome of the study sample

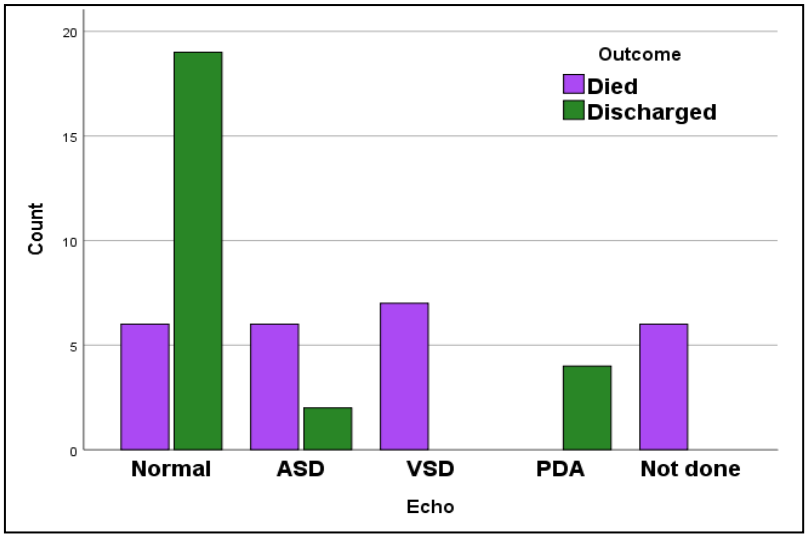

Figure 13: Congenital heart disease

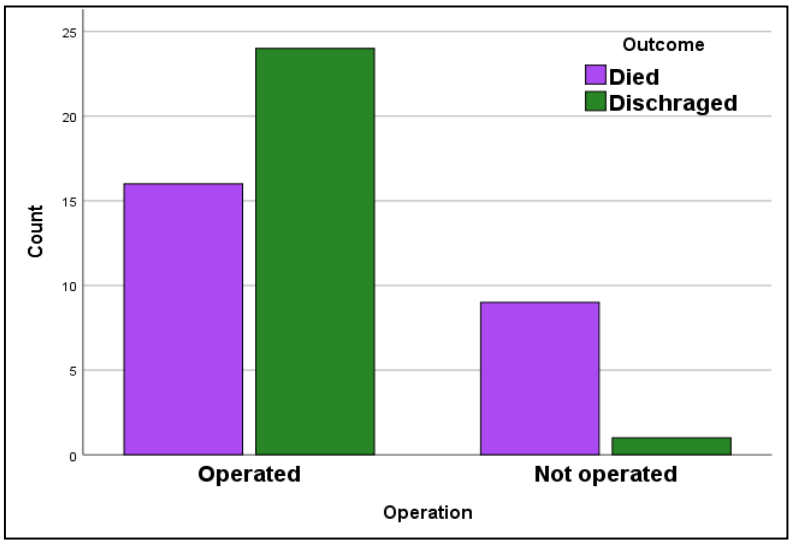

Figure 14: Operative management and outcome of the study sample

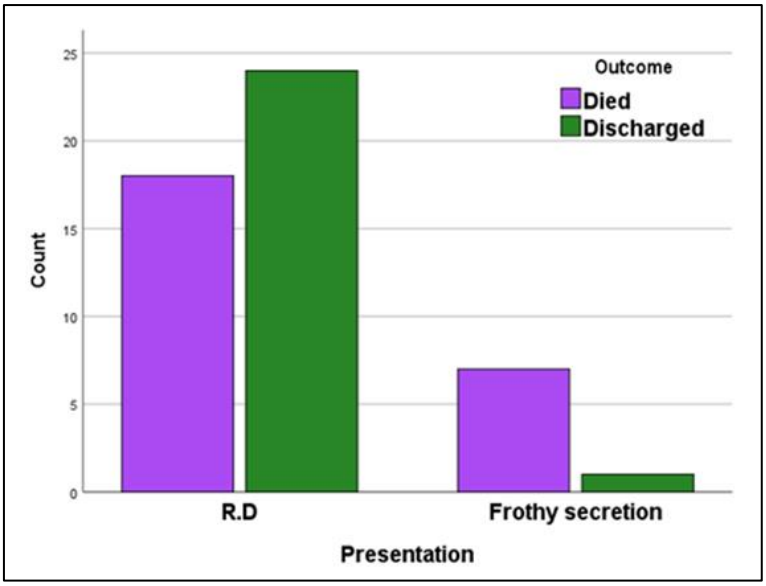

Figure 15: Presentations and outcome of the study sample.

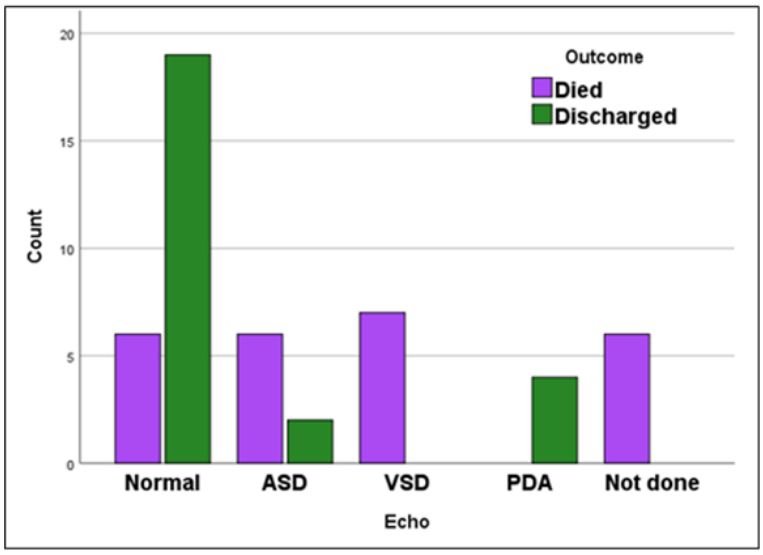

Figure 16: Echo results and outcome of the study sample .

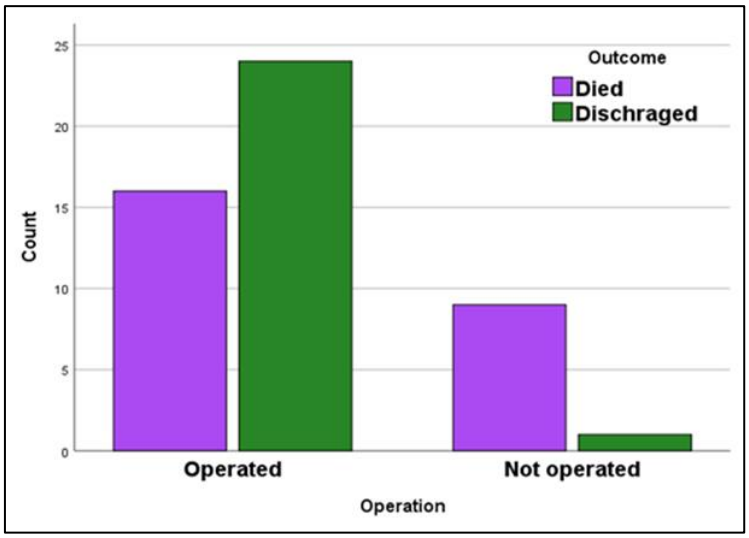

Figure 17: Operative management and outcome of the study sample.

DISCUSSION

Respiratory distress is a common problem during the neonatal period with considerable mortality as it accounts for nearly half of all deaths. It is a heterogeneous group of illnesses with varying prevalence, underlying etiology, clinical course and outcome; many of the underlying causes of respiratory distress in a newborn are unique to this age group. Prompt investigation to ascertain the underlying diagnosis and appropriate subsequent management is important to improve outcome [56]. Respiratory distress may be due to either medical or surgical cause [57]. Neonatal respiratory disorders account for most admissions to Neonatal intensive care units in the immediate newborn periods [58]. Respiratory distress is a major problem of ill -health in the newborn and reported in (20 -50%) of admissions in the neonatal care units in developing countries [59, 60].

Initial and careful assessment of the infant in respiratory distress is very important to identify life threatening conditions that require prompt support, such as inadequate or obstructed airway (gasping, choking, stridor), apnea or poor respiratory efforts, cyanosis, and circulatory collapse (bradycardia, hypotension, poor perfusion).

Gender distribution of the sample of this study was predominantly male as (70%) of them were males, and (30%) were females. This finding is similar to many other studies, for example a study describing TA as a rare congenital airway malformation, characterized by complete or partial absence of the trachea, frequently associated with carino- esophageal fistula, bronchoesophageal fistula, or tracheo - esophageal fistula (TOF). It was found that, the incidence is less than (1:50,000), with a male preponderance and a high correlation with isolated or multiple congenital malformations, usually part of the "VACTERL" association (vertebral defects, anal atresia, TOF with esophageal atresia and radial or renal dysplasia, plus cardiovascular and limb defects), and the "TARCD" association (TA or tracheal atresia, radial ray defects, complex congenital cardiac abnormalities, and duodenal atresia) [61].

Despite advances in treatment during the last six decades, mortality of TOF patients continue to be high, the sample of this study showed that TOF cases were in total (26) cases of them (17) (65.4%) died and (9) survived, diaphragmatic hernia cases were (20), of them eight cases (40%) died (12) (60%) discharged, all cases of lung emphysema discharged. This is mortality for RD due to congenital malformation which might require prompt diagnosis and surgical intervention. This might be deficient in our setting as the hospital suffers from reduced facility for diagnosis and treatment of such urgent conditions. Other medical caused of RD was reported by previous study as a common neonatal problem with significant morbidity and mortality. They concluded that medical caused are treatable with success which could be due to transient tachypnea of newborn which is self - limiting disease and need only observation. Prevalence of hyaline membrane disease can be reduced by prenatal administration of corticosteroids and similarly cases due to meconium aspiration syndrome could be reduced by improved antepartum, intrapartum care and the post -delivery resuscitation of newborn [62].

A previous study found the total of 4168 cases was identified with diagnosis of neonatal RD. the overall in - hospital mortality was (9%). Univariate analysis revealed lower survival in patients with associated acute respiratory distress syndrome, ventricular septal defect (VSD), birth weight (BW) < 1500 g, gestational age (GA), time of operation within 24 h of admission, coexisting renal anomaly, imperforate anus, African American race, and lowest economic status. Multivariate logistic regression identified BW < 1500 g (odds ratio [OR] = 4.5, P < 0.001), operation within 24 h (OR = 6.9, P < 0.001), GA <28 wk (OR = 2.2, P < 0.030), and presence of VSD (OR = 3.8, P < 0.001) as independent predictors of in -hospital mortality. Children's general hospital and children's unit in a general hospital were found to have a lower mortality rate compared with not identified as a children's hospital after excluding immediate transfers (P = 0.008) [63].

Diagnosis of the cases of this study showed that TOF was the most frequent which occurred in (52%) of the cases, followed by diaphragmatic hernia in (40%), then lung emphysema in (8%).Clinical presentation of the cases was mainly with respiratory distress which occurred in (84%) of the sample, while (16%) presented with frothy secretion. These results are in accordance to previous research which concluded that; congenital anomalies within the airways and lungs may require surgical correction. The most common problems are congenital diaphragmatic hernia (CDH), congenital pulmonary airway malformation (CPAM), Tracheo - oesophageal fistula (TOF) [64].

In a previous study the clinical presentation of TOF varies according to the presence or absence of EA. Infants with TOF and EA usually become symptomatic immediately after birth, with drooling, choking and respiratory distress during swallowing. In addition, the air passing through the fistula leads to gastric distension and subsequent reflux of gastric contents through the same TOF, resulting in aspiration pneumonia. Instead, patients with isolated TOF, also defined H - type, may become early symptomatic at the time of feeding if the defect is large, or exhibit belatedly symptoms if the defects is small [65].

In Kingdom of Saudi Arabia a study done from January 1998 - December 2013 in which (435) infants and children with the diagnosis of esophageal atresia

with or without TOF were treated. Among these, (23) (5.3%) had isolated TEF. There were (11) males and (12) females. Their age at presentation ranged from (5) days to (3) years and (7) months but the majority (90%) were diagnosed during their first year of life. Their clinical presentation included: chocking and coughing during feeds in (12) (52.2%), recurrent chest infection in (16) (69.6%) and cyanosis in (10) (43.5%). One presented with abdominal distension also. The diagnosis was made using esophagogram. In (11) (47.8%), a single study confirmed the diagnosis, (8) (34.8%) required two studies while (4) (17.4%) required three studies. Nineteen (82.6%) had preoperative bronchoscopy and in (13) (56.5%), a catheter was used to cannulate the fistula. All were operated through a right cervical incision except one who underwent thoracoscopic ligation and division of the fistula. In one, the fistula was only transfixed and tied without being divided. This patient developed a recurrent fistula. Two patients developed postoperative stridor secondary to recurrent laryngeal nerve palsy. In both of them, there was complete recovery [66].

Congenital lobar emphysema is commonly observed in left upper lobe (43%), followed by right middle lobe (32%) and right upper lobe (21%) involvement. Lower lobe involvement (2%) is the rarest form. In the literature, more than one lobe and bilateral involvement have been described. Nearly half of patients are symptomatic at birth, while the other half mostly develops symptoms in the first 6 months of life. The affected lobe is overinflated, and ventilation and perfusion are impaired in the overinflated lobe. With progressive over inflation, compression occurs in the adjacent organs. Thus, ventilation and perfusion are impaired in these parts of the lung parenchyma, which leads to progressive respiratory failure. Retractions, wheezing, cyanosis, and difficulty in feeding can be observed. In infancy, wheezing, chronic cough, and recurrent respiratory tract infections can be seen. In the literature, it was reported that a (15) - year - old boy was diagnosed with CLE without respiratory symptoms. Some patients can be mistakenly diagnosed with pneumothorax and pneumonia instead of CLE in later life [67].

Infants with CDH often present in the neonatal period with severe respiratory distress. Presentation after infancy, occurring in (5%) - (10%) of affected individuals, includes respiratory distress, such as from pleural effusion due to entrapment of the bowel in the chest, or gastrointestinal distress, such as abdominal pain from chronic or intermittent intestinal obstruction. About (1%) of individuals are completely asymptomatic and the defect is discovered incidentally on imaging studies. At least (10%) of individuals reherniate following initial surgical repair; the risk is considerably greater among those whose hernia repair required a prosthetic patch [68].

CONCLUSION

A sample of (50) children were studied, they were admitted as cases of respiratory distress due to congenital anomalies, the age varied from few days to years, neonates represented (44%) of the sample, male children formed (70%) of the sample and (30%) females. Most of the cases were from outside Benghazi. Just more than half of the sample were born as normal vaginal delivery while (48%) via C/S, (86%) of them were full term. TOF was the most frequent cause of respiratory distress among this sample, followed by diaphragmatic hernia, few cases had lung emphysema. Clinically the cases were presented with respiratory distress in (84%) of them, or frothy secretions in (16%). USS was normal in (88%), echo was normal in (50%), revealed atrial septal defect in (16%) of the sample, ventricular septal defect in (14%), patent ductus arteriosus in (8%) and not done to (12%) of the sample. The underlying congenital anomalies was operated in as (80%) of the sample and not operated the remaining (20%). The final outcome was Death in half of the cases and discharged in the other half.

Predicting cases outcome was not based on variables such as gender, mode of delivery, city of residence which showed no significant association with cases outcome. Poor outcome predictors of the current study were:

- **Diagnosis:** TOF cases were in total (26) cases of them (17) (65.4%) died and (9) survived, diaphragmatic hernia cases were (20), of them eight cases (40%) died (12) (60%) discharged, all cases of lung emphysema discharged.
- **Birth Maturity:** All preterm babies of this study showed poor outcome as all of them died (seven cases) and those who were term were in total (43) of them (25) survived and (18) died.
- **Clinical Presentations:** Cases were either presented with respiratory distress or frothy secretions, frothy secretions presentation was predictor for worse outcome as in total (8) of them (7) died. Cases who were presented with respiratory distress were in total (42) of them (18) died and (25) discharged, while cases who.
- **Radiological Investigations:** Cases with normal USS showed better outcome (44 cases of them 19 died and 25 survived) while it was not done to six cases and all of died which could be due to inability to do it or immediate death of the child.
- **Echo:** Cases who showed normal echo were (25) cases of them (19) discharged and only (6) died, cases with ASD were (8) of them (6) died, (7) cases with VSD all of them died, PDA associated with good outcome as they were four cases all of them discharged, and echo was not done to (6) cases and all of them had died.

- **Surgical Intervention:** Operation for congenital anomalies was done to (40) cases of them 16 died and (24) discharged, and not done to (10) cases of them (9) died and one case discharged.

RECOMMENDATIONS

- Infants with RD usually referred as multiple aetiologies case, medical and surgical causes will be accused, the affected children need to be evaluated by a multidisciplinary team, including pulmonology, otolaryngology, gastroenterology and speech pathology.
- Medical causes can be prevented by improved antepartum, intrapartum care and the post - delivery resuscitation of newborn.
- Clinicians need to be aware of congenital airway malformations, as prenatal diagnosis is mandatory to opt for pregnancy termination or plan for a multidisciplinary management of delivery.
- Conversely, whenever prenatal diagnosis is impossible, difficulties upon endotracheal intubation and several clinical features should immediately alert the clinician of a possible airway malformation as there is little time for emergency management of the airway, including intubation through esophageal fistula or tracheostomy.

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