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Original Research Article

Prenatal and neonatal diagnosis of congenital lung lesions and their management

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ABSTRACT

Background: The therapeutic strategy for congenital lung lesions remains controversial. The majority of the patients with congenital lung lesions do not present with complications. Some authors report that there should be no indication of surgical intervention for babies with no symptoms. Conversely, other authors recommend elective surgery, because the wait-and-see strategy might only increase the risk of abscess formation or the development of severe lung infections. The aims of the study was to evaluate cases with congenital lung lesions presented to pediatric Surgical department with an objective of assessing age and sex distribution, clinical features, diagnosis, management and outcomes.
Methods: Prospective and retrospective study in Department of Pediatric Surgery, all cases of congenital lung lesions diagnosed antenatally and in neonatal period. Total 45 Patients were included.
Results: Postoperative complications seen in 8 patients with morbidity about 25%. The surgical outcome is favorable, with manageable complications. Surgical intervention in asymptomatic patients is suggested. As the incidence of symptomatic, patients is highest between 1 to 6 months. Recommend elective surgery at 1 month of age. Surgery in asymptomatic patients is less invasive and in the infancy it is safer and more beneficial.
Conclusions: Neonates, infants, and children presenting with respiratory distress due to these developmental anomalies may require urgent surgical intervention. Plain X-ray chest and CECT scan of the thorax are usually sufficient for diagnosis and planning of treatment. Lobectomy is the procedure of choice.

Keywords: Respiratory distress, Lobectomy, Neonatal diagnosis

INTRODUCTION

The therapeutic strategy for congenital lung lesions remains controversial. It is well known that, the majority of the patients with congenital lung lesions do not present with complications, which include respiratory failure, pneumonia and lung abscess, immediately after birth or for the first few months of life. Some authors report that there should be no indication of surgical intervention for babies with no symptoms.[1,2] Conversely, other authors recommend elective surgery, even on asymptomatic babies with congenital lung lesions because the spontaneous regression of the cystic lesion after birth is reported to be unlikely, and because the wait-and-see strategy might only increase the risk of abscess formation or the development of severe lung infections.[3,4] Such infectious episodes might also increase the risk of Intraoperative and postoperative complications in surgical interventions.

It has also been reported that the presence of the congenital lung lesion could negatively affect compensatory growth of normal lung.[5] We assessed the outcome of our 6 year experience of congenital lung lesions in Niloufer hospital, Hyderabad with a focus on the clinical feature, diagnosis, management and safety of lung resections.

METHODS

This is a prospective and retrospective study includes cases of congenital lung lesions diagnosed antenatally and neonatally presenting to Pediatric Surgery Department in Niloufer Hospital from January 2010 to October 2014 (retrospective) and November 2014 to November 2016 (prospective).

Inclusion criteria

All cases of congenital lung lesions diagnosed antenatally and in neonatal period.

Exclusion criteria

Cases with associated malformations.

All demographic details were noted. Common clinical presentations and associated cardiac malformations were noted. All patients had done with chest radiography. Diagnostic studies included CT scan, MRI, arteriography and bronchoscopy.

Type of resection

Emergency resections were performed (CCAM 25; CLE 17; Bronchogenic cyst 3). Surgical procedures included lobectomies, sequestrectomies, omediastinal mass excision and wedge resection for a bronchogenic cyst. Surgical access was by right or left thoracotomy in most patients. All resected specimens were submitted for histopathology.

In congenital cystic adenomatoid malformation (n=25) surgical treatment for congenital cystic adenomatoid malformation consisted of lobectomy and wedge resection of the lobe for extensive CCAM, but developed a prolonged air leak postoperatively and required a completion right lower lobectomy, sent for histopathological examination.

Cases of lobar hyperinflation secondary to intraluminal or extrinsic obstruction were excluded. Neonates who had respiratory distress and marked mediastinal shift, requiring an emergency lobectomy. Patient required ventilation preoperatively. Bronchoscopy was performed in children to rule out intrinsic obstruction where ever required. Surgical treatment consisted of right upper lobectomy, right middle lobectomy, and left upper lobectomy. None of these patients had pulmonary hypoplasia.

Following the initial diagnosis on antenatal ultrasound, the in utero course of these patients evolved as described. In ten patients, antenatal ultrasound at the end of pregnancy was considered to demonstrate resolution of the anomaly. Infants with CCAM were shown to have a persistent lesion, which was not demonstrable on radiographs but could be depicted on postpartum multi-slice CT scans. The sequestration patients were shown to have persistent lesions on both chest radiographs and CT scan.

In some cases serial antenatal sonograms towards the end of pregnancy demonstrated spontaneous resolution of the malformation, which was confirmed by postpartum ultrasound, radiographs and CT scans. Serial therapeutic amniocenteses were performed in three fetuses with polyhydramnios. Re-accumulation of fluid occurred in one foetus while the others improved. Foetuses with malformation remained stable and neither progressed nor regressed. There were no associated mass effects with these lesions. Patient diagnosed to have CCAM on antenatal ultrasound, was shown to have an enteric cyst on postoperative histopathology.

There was no evidence of progression of hydrops, mediastinal shift, or cyst enlargement in any of the foetuses on sequential scanning. No foetuses in this series had other structural anomalies detected on ultrasound. No pregnancy was terminated. There were no instances of foetal demise.

Total number of patients included was 45. Patients were followed up at 1st week and monthly thereafter for two years. During two year follow up recurrence of the disease was monitored.

RESULTS

The clinical study of prenatal and neonatal diagnosis of congenital lung lesions and their management was conducted in the Department of Pediatric surgery, Niloufer Hospital, Hyderabad, during a period six years from January 2010 to till date (December 2016).

The total no of patients admitted were 31600 during this period. Congenital lung lesions were 45 comprising 0.15% of total admissions.

Table 1: Demographic distribution.

Demographic distribution	Number of cases	Percentage (%)
Male	26	57.8
Female	19	42.2
Male: female	1.37:1	
Age distribution		
NB- 30 days	10	22.23
1 month-6 months	17	37.77
6 months-1 year	10	22.23
Above 1 year	8	17.77

The highest incidence of the disease observed in age group 1–6 months (37.77%) and 37 cases (82.23%) presented under 1 year of age. Majority of the patient (60%) presented with acute respiratory Infection.

A total of 45 congenital lung lesions were treated. 26 patients had cysts in Rt. lung and 17 patients had in the left lung.

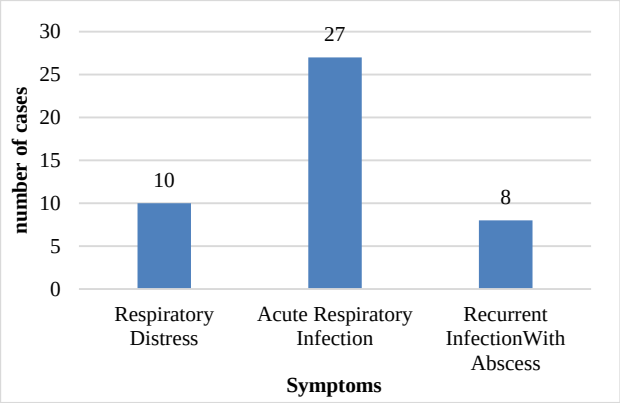

Figure 1: Common symptoms in present study.

Table 2: Preoperative diagnosis of congenital lung lesions.

Diagnosis	Number of cases	Percentage (%)
Congenital cystic adenomatoid malformations	25	55.56
Congenital lobar emphysemas	17	37.77
Bronchogenic cysts	3	6.67

Most common congenital lung lesion in this study is CCAM Comprising 55.56%, followed by CLE 37.77% and BC 6.67%.

CCAM most commonly affected right lower lobe followed by left lower lobe. CLE most commonly affected right middle lobe followed by left upper lobe.

Table 3: Lobar distribution of congenital lung lesions.

Diagnosis	RUL	RML	RLL	LUL	LML	LLL
Congenital cystic adenomatoid malformations	1	3	10	2	1	8
Congenital lobar emphysemas	1	9	0	4	2	1
Bronchogenic cysts	1	0	0	1	0	0

Table 4: Operating time and blood loss in present study.

Age	Range of operation time in mins	Mean operation time in mins
Neonates	40-60	50.5
1 to 6 months	45-80	52.9
6 t0 12 months	50-80	57.8
Above 1 year	70-120	81.2
Intra operative blood loss	**Range of blood loss in ml**	**Mean blood loss in ml**
Neonates	40-50	43.5
1 to 6 months	45-60	45.6
6 t0 12 months	50-80	48.3
Above 1 year	70-120	72.3

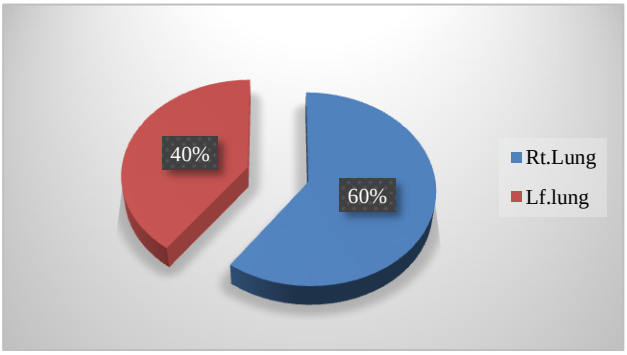

Figure 2: Location of the lesion in present study.

Operating time for neonates ranging from 40-60 mins, for infants 45-80 mins and for children aged above 1 year is 70-120 min. Blood loss for neonates and infants is range between 40–80 ml and for children above 1 yr is above 70-120 ml.

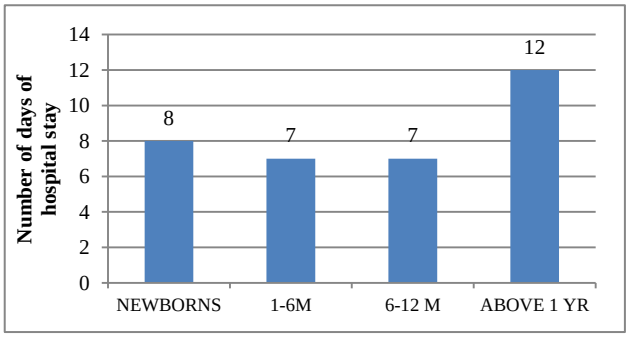

Figure 3: Hospital stay in present study.

The mean hospital stay was 10 to 14 days for children above 1 year, for newborns it is 8 days and for infants 7 days.

Postoperative complications commonly seen in older children presented with lung abscess. Intraoperative bleeding in 4 cases and postoperative air leak in 2 cases.

Mortality rate in our study is 3 [new borns with CCAM (6.67%)]. Patients were followed up at 1st week and monthly thereafter for two years. All patients have good pulmonary function and growth. During two year follow up no cases had recurrence of the disease.

DISCUSSION

The surgical intervention strategies for congenital lung lesions especially for patients without any Symptoms remain controversial. It has been said that indications for lung resection in patients with congenital lung lesions should be determined considering the risk of infectious complications, ongoing respiratory distress and malignant transformation.[6]

It is impossible to predict when such complications occur during early life. The general condition and body weight should also be taken into account to minimize the incidence of perioperative complications. All these factors make it difficult to determine the most appropriate timing of surgical Intervention for asymptomatic patients. Recently, Baird et al reported a management strategy for congenital lung malformations which utilized the grading of recommendations assessment, Development and evaluation process (GRADE).[6]

This review stated that prophylactic resection should not be recommended for asymptomatic children with congenital lung lesions. Kotecha also recommended medical observation for asymptomatic children with Congenital cystic adenomatoid malformations (CCAM) and only promoted surgery for Children who were identified as being at risk of developing future symptoms.[7] Peters et al reported that 20% of surgeons in the UK and Ireland never resects an asymptomatic lesion.[8]

Ng et al reported that 5% of asymptomatic infants developed symptoms associated with CCAM after a median follow-up period of 5 years.[9] Conversely, some authors recommend elective surgery in asymptomatic Children with Congenital lung lesions out of consideration for the risk of emergency surgery to treat Sudden onset Complications and the disturbance of compensatory growth of normal lung.[10] Some authors have reported that the risk of infection for congenital lung lesions has been estimated to range between 10% and 30% within the first year of life. Pelizzo et al reported that early signs of inflammation could be present even in asymptomatic infants who underwent surgery within the first 3 months of life.[11]

Marshall et al concluded that surgical intervention in the asymptomatic infant is associated with a shorter length of stay, a trend toward fewer complications, and decreased medical cost in comparison to intervening after the development of symptoms.[12] Laberge et al recommended that lung resection should be performed at 3 to 6 months of life at the latest, so that compensatory lung growth could occur.[5] Komori et al reported that patients who were younger than 1 year at the time of surgery showed significantly better pulmonary function than patients who were older than 1 year at the time of surgery.[14] Some authors have also suggested that lung resection should be performed before the age of 1 year.[15] Total no of patients included in our study.

Of the 45 patients 26 were males and 19 were females. With male to female ratio of 1.37:1.

Table 5: Comparison of present study with other studies.

	Tsai et al[16]	Furukawa et al[17]	Present study
Male:female	0.8:1	-	1.37:1
Age (range in months)	1.0-13.3	-	Newborn- 132
Mean age	2.5 ± 2.4		14.2
Mode of presentation			
Respiratory distress		1 (7.69%)	10 (22.3%)
Acute respiratory infection		8 (61.53%)	27 (60%)
Aecurrent infection with abscess		3 (37.5%)	8 (17.7%)
Side of lung			
Right lung	52 (50%)		27 (57.77)
Left lung	52 (50%)		18 (42.33)
Mean operation time			
Duration average (min)	167 (asymptomatic)		50 (asymptomatic)
	275 (symptomatic)		80 (symptomatic)
Duration range (min)	40-260		60-120
Length of hosp. stay average (d)	3.0 ± 1.1		7
Range (d)	1-9		7-14

Sex incidence in our study (M:F- 1.37:1) does not correlates with study done by Tsai et al.[16] Majority of the patients were in the age group 1 to 6 months in present study.

Age incidence in our study (mean age 14.2 months) is not comparable with other studies. The common mode of presentation was acute respiratory infection seen in 60% of the patients. The most common symptomatology in our study is acute respiratory infection 60% comparable with the study done by Furukawa et al.[17]

The presence and location was confirmed by CECT Chest. Among 45 patients, 27 patients had cysts in the Right lobe and 18 had cysts in the Left lobe.

The mean operation time was longer in patients presented late with complications.

The mean hospital stay was 7 days for patient presented early without symptoms and 10.8 days for patients presented late with symptoms. Postoperative complications seen in 8 patients with morbidity about 25%. Recurrence in our study was 0% which is comparable with other studies.

CONCLUSION

In conclusion, the analysis of the patients in our series suggested that these anomalies are comparatively rare. Neonates, infants, and children presenting with respiratory distress due to these developmental anomalies may require urgent surgical intervention. Plain x-ray chest and CECT scan of the thorax are usually sufficient for diagnosis and planning of treatment. The surgical outcome is favorable, with manageable complications. Lobectomy is the procedure of choice in our series. Our findings indicate that mortality is higher in neonates. In addition to the initial stabilization, resection of lesion and careful postoperative care is necessary for improvement of the respiratory status in neonates and infants. Progressively increasing respiratory distress that shows no response to conservative treatment is an absolute indication for surgery, regardless of age; In our study morbidity is high; duration of surgery is long, postoperative complications are more and hospital stay is longer in patients presented at later age with complications because of more invasiveness of the surgery.

Surgical intervention in asymptomatic patients is therefore suggested. As the incidence of symptomatic Patients is highest between 1 to 6 months of age in our study we recommend elective surgery at 1 month of age. Surgery in asymptomatic patients is less invasive and in the infancy it is safer and more beneficial.

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