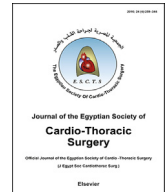




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Surgical management of congenital lobar emphysema: A 15 years experience in a tertiary thoracic surgery unit

Essam Elbadry Hashim Mohamed ^{a, *}, Ayman Mohamed Mohamed Abdel Ghaffar ^a, Khaled Mohamed Abdel-Aal ^a, Abdelhady Ahmed Helmy ^b, Mostafa Ashry ^c

^a Cardiothoracic Surgery Department, Sohag Faculty of Medicine and University Hospital, Sohag University, PO 82524, Sohag, Egypt

^b Anesthesia and Intensive Care, Sohag Faculty of Medicine and University Hospital, Sohag, Egypt

^c Pediatrics and Neonatology, Sohag Faculty of Medicine and University Hospital, Sohag, Egypt

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ABSTRACT

Background: congenital lobar emphysema is a rare cause of respiratory distress in neonates and infants. It was treated by lobectomy since 1943.

The aim of this study was to present our experience in the diagnosis and surgical treatment of congenital lobar emphysema cases and highlight the effect of multi-disciplinary team work on the outcome.

Methods: We retrospectively analyzed data of patients operated for congenital lobar emphysema at cardiothoracic surgery department, Sohag university hospital over the period of 15 years starting from January 2000 and ending in December 2015.

Results: Total number of cases was fifty three. Males were 39 (74%) while females were 14 (26%). Mean age was 12 weeks (3 weeks - 15 months). Six patients (11%) had associated cardiac anomalies. Main clinical presentations were acute respiratory distress and recurrent chest infection. Left upper lobe was affected in 35 cases (66%), right upper lobe in 10 cases (19%) and right middle lobe in 8 cases (15%). All cases were subjected to anatomical lobectomy by thoracotomy. One patient was subjected to patent ductus arteriosus ligation simultaneously with left upper lobectomy at the same operative setting. Post operative complications were reported in 8 cases (15%) and one patient of them died from sepsis and respiratory failure (1.9%). Postoperatively, all surviving patients were followed up for one year in the outpatient clinic.

Conclusions: congenital lobar emphysema is a life threatening disease. Early diagnosis and surgical management by a multi-disciplinary team is the golden key for favorable and safe outcome.

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List of abbreviations: CLE, Congenital lobar emphysema; UL, Left upper lobe; RML, Right middle lobe; RUL, Right upper lobe; CT, computed tomography; CXR, chest X ray; PO, post operative; PDA, patent ductus arteriosus; VSD, ventricular septal defect; ASD, atrial septal defect; OR, operation room; ICU, intensive care unit; Fig., figure; CPAM, congenital pulmonary airway malformation.

* Corresponding author. Tel.: +20- 01093939202; fax: +20-0934609304.

E-mail addresses: essam_hashem@med.sohag.edu.eg (E.E.H. Mohamed), aymangf2001@yahoo.com (A.M. Mohamed Abdel Ghaffar), km.abdelaal@yahoo.com (K.M. Abdel-Aal), 71abdelhady@gmail.com (A.A. Helmy), mostafashry@yahoo.com (M. Ashry).

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1. Introduction

Congenital lobar emphysema (CLE) is a rare developmental malformation of the lung. Its incidence being nearly 1 per 25000 births [1]. This disease entity was precisely described for the first time in 1932 by Doctor Nelson [2]. This lung abnormality often presents in the neonatal period and early infancy, so sometimes termed infantile lobar emphysema [3,4]. It is characterized by over-inflation of one or more lung lobes which is due to localized air trapping that compresses the ipsilateral and contralateral normal lungs. The cause of CLE is not yet fully understood; bronchial abnormalities [5] or alveolar defects [6] have been reported as etiological factors. The most frequently affected lobe is the left upper lobe (LUL), followed by the middle and then the right upper lobe (RUL) [7]. Males are more commonly affected with a male to female ratio is 3:1 [8]. The most common clinical presentation is neonatal acute respiratory distress. Dyspnea, tachypnea, wheezing, cough, and cyanosis are among the most common presenting symptoms. Symptoms may be exaggerated by feeding or crying [9,10]. The time of onset and severity of symptoms varies depending on the degree of hyperinflation. In some infants, progressive respiratory distress develops rapidly, while others have a more gradual and insidious onset, 50% presenting by one month of age [11,12]. On the other hand, there is a rare adult presentation [13]. Some authors reported CLE as a rare cause of hypertension [14]. Physical findings include hyper-resonant percussion, diminished breath sounds on the affected side and mediastinal shift which manifested by displacement of the cardiac point of maximal impulse. The chest may appear hype-expanded with limited respiratory excursion. The infants may present with recurrent respiratory infections or poor feeding with failure to thrive [10]. Other congenital anomalies may be associated with CLE. Cardiovascular anomalies are the most common which represent up to 20% of the associated anomalies [15,16]. The diagnosis is suspected by clinical examination and chest X-ray imaging which can be confirmed by computed tomography (CT) of chest [17]. The traditional treatment of CLE is lobectomy of the involved lobe or lobes [1,17–19]. Some authors reported conservative treatment for patients with mild symptoms [10,20,21]. The first curative lobectomy for CLE was performed on a four-year-old girl in 1943 by Gross and Lewis [12], then many reports of successful surgical treatment have been published [1,19,22].

The aim of this study was to present our experience in the surgical treatment of children with CLE at Cardiothoracic surgery department, Sohag university hospital over 15- year period. This was done by reviewing the clinical presentation, radiographic findings, the surgical approach and the post operative outcomes of these children. In addition, the aim was to highlight the effect of multi-disciplinary team works on the outcome.

2. Patients and methods

This is a retrospective study performed at Sohag university hospital in Egypt. The medical records of all children who were subjected to surgical treatment of CLE in the period between January 2000 and December 2015 at Cardiothoracic surgery department were reviewed.

The following data were collected and analyzed:

Pre-operative data included age at time of surgery, gender, clinical picture, chest X ray findings (Fig. 1), CT chest findings (Fig. 2A,B&C), Bronchoscopy findings (when performed), affected lobes and presence of associated congenital malformations.

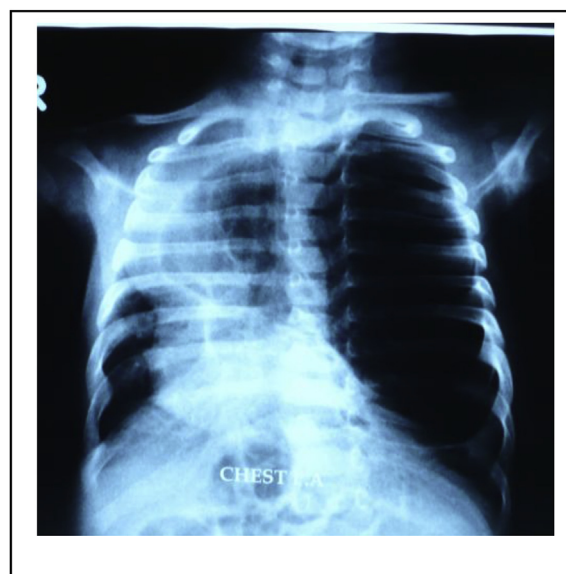


Fig. 1. Chest X ray of 3 months old infant showing CLE of LUL.

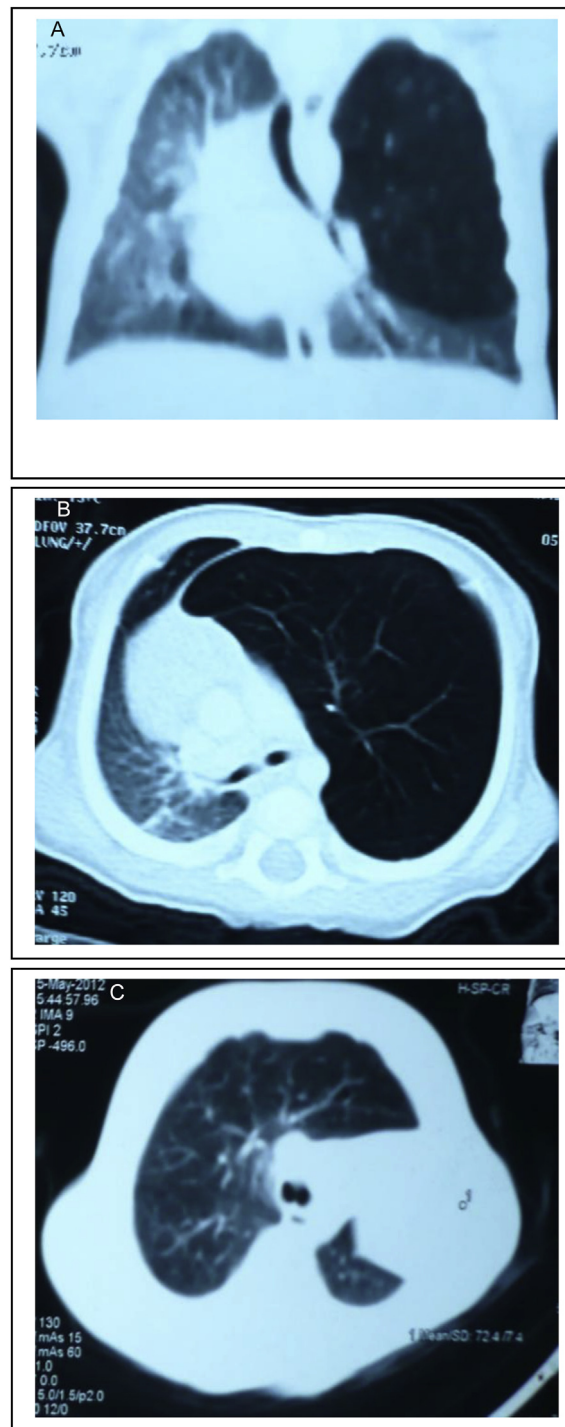


Fig. 2. A: coronal view of CT chest in a 3 months old infant showing CLE of LUL. B: CT scan of the chest, axial view in 3 months old infant, shows CLE of LUL. C: CT scan of the chest, axial view in 4 months old infant, shows CLE of RUL.

Operative data included approach, type of surgery performed, operative findings, operative morbidity and mortality.

The anesthesia teams in collaboration with the surgeons exerted a special effort to prevent cardiopulmonary compromise during induction of anesthesia as the emphysematous lobe is at risk of more hyperinflation and more compression of the mediastinum and adjacent normal lung tissue. The patient was sterilized and draped under sedation, and then induction started while the surgeon was standby for thoracotomy which was performed rapidly to allow the

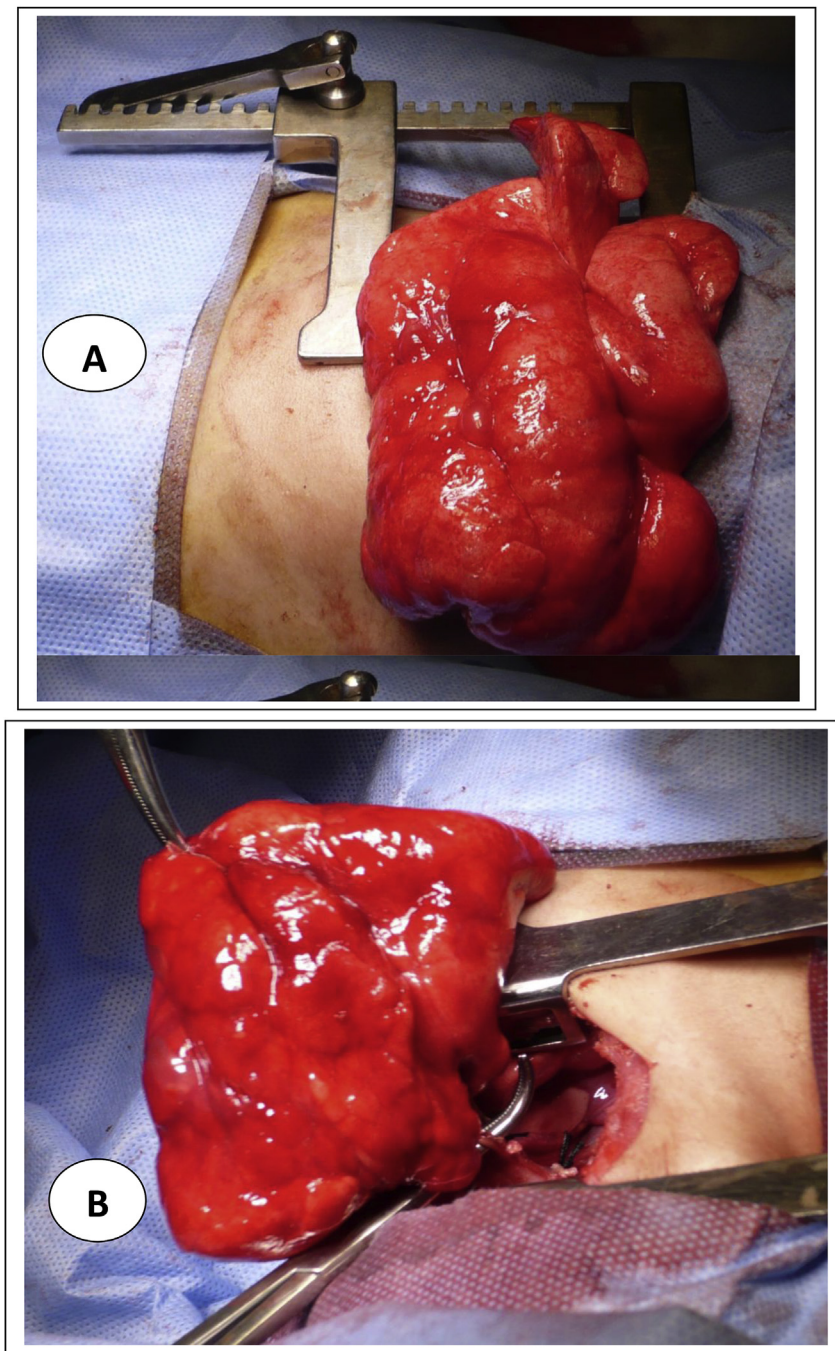


Fig. 3. Intra-operative images of left upper lobectomy for CLE in 3 months old boy. **A:** herniation of the emphysematous lobe outside of the wound just after opening of the chest. **B:** showing anatomical resection of the left upper lobe.

emphysematous lobe to be herniated outside the wound (Fig. 3-A) which is a characteristic operative finding for CLE. Lobectomy had been performed in all cases (Fig. 3-B). After resection, the remaining lobe, which was compressed, expand to fill the chest. One Chest drain was inserted and the thoracotomy was closed in layers. The resected lobe sent for histopathology (Fig. 4). After completion of the operation, patients were routinely extubated in the OR and transferred to pediatric ICU for monitoring. Post operative chest X ray (Fig. 5) was usually performed in the next morning and then when needed. After confirmation of absence of air leak with fully expanded lung and no bleeding, the chest drain was removed. Postoperative morbidity and mortality were reported and analyzed. All patients were followed in hospital and in outpatient clinic by thoracic surgeons together with pediatricians. All the surviving cases were followed up for one

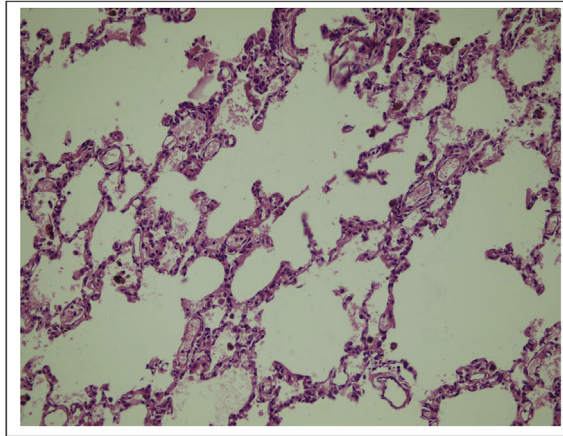


Fig. 4. Microscopic picture of CLE show Over-distention and dilatation of alveolar spaces with focal rupture of alveolar septa.



Fig. 5. CXR PA view showing post operative status (day one) post left upper lobectomy for CLE in a 3 months old boy.



Fig. 6. Follow up CXR of an infant 6 months post left upper lobectomy for CLE.

year after discharge. Follow up was based on Clinical and radiographic findings (Fig. 6) on regular visits at outpatient clinic.

2.1. Statistical analysis section

Values are presented as numbers(%) or mean (range), as indicated.

3. Results

At the Cardiothoracic surgery department of Sohag university hospital in the period from January 2000 to December 2015, fifty three cases were subjected to surgical treatment for CLE. Their ages at surgery ranged from three weeks to fifteen months (mean age 12 weeks). The most commonly affected lobe was the LUL (35 cases) followed by RUL (10 cases) then RML (8 cases) (Diagram 1). Thirty nine (74%) patients were males and fourteen (26%) patients were females (Diagram 2). Symptoms of respiratory distress by different degrees were the most common presenting symptom followed by manifestations of repeated pulmonary infections. All cases were diagnosed initially by plain CXR PA view which showed hyperinflation of the affected lobe herniating across the midline, atelectasis of underlying and contralateral normal lung with mediastinal shift. Forty cases (75%) were subjected to CT chest examination before surgery to confirm the diagnosis. Pre-operative bronchoscopy was performed in 7 cases (13%), Rigid bronchoscopy was performed in 5 cases, all of them were over 8 months of age while fiberoptic bronchoscopy performed in 2 cases. No specific findings were reported in any of them. Congenital cardiac malformations were reported in six cases in our series, two cases of isolated PDA, three cases of ASD and one case of VSD. Pectus excavatum was reported in one case. PDA ligation was performed simultaneously with left upper lobectomy in one case, the other case of PDA was associated with CLE of the right upper lobe, right upper lobectomy was performed and PDA was later on closed in another session. Cases of ASD and VSD managed for CLE and the management of cardiac anomalies was postponed to be managed later on. In all cases, the emphysematous lobe was subjected to lobectomy through muscle sparing postero-lateral thoracotomy. After surgery, all of the patients recovered from anesthesia and weaned from mechanical ventilation in the operating room except for one case transferred to ICU on mechanical ventilation, this patient was already on pre-operative ventilation because of pneumonia and respiratory failure. Postoperative complications reported in 8 cases (15%), wound infection had happened in 2 cases, one of them was mild and managed by frequent dressing and manipulations of antibiotics, but in one case the infection was heavy and associated with encysted empyema which require re-entry to the OR where wash

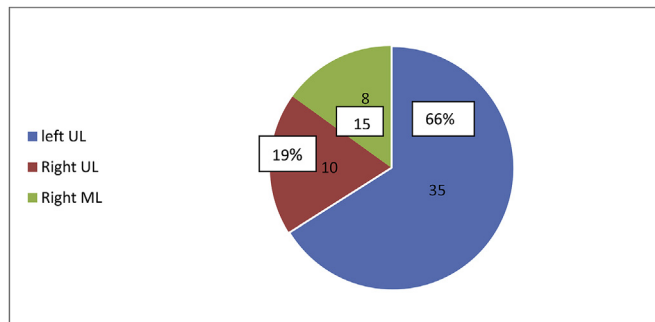


Diagram 1. Anatomical distribution of CLE cases (Total No. : 53).

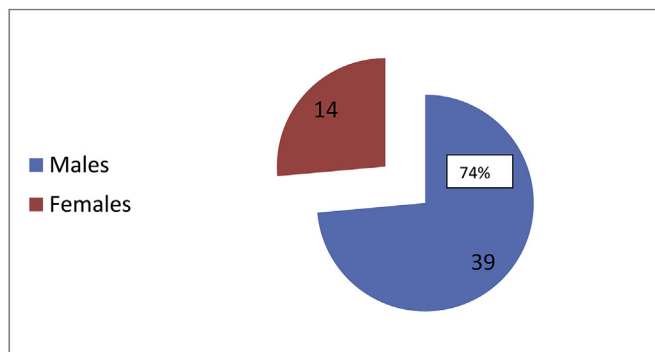


Diagram 2. Gender distribution of CLE cases (Total No. :53).

Table 1

Analysis of data of patients with CLE treated by Lobectomy.

Total Number	53		
Gender	Males	39	74%
	Females	14	26%
Affected Lobe	LUL	35	66%
	RUL	10	19%
	RML	8	15%
Age	Youngest	3W	
	Oldest	15M	
Bronchoscopy	Total	7	13%
	Rigid	5	
	Fiber-Optic	2	
All Associated Anomalies	Total	7	13%
	Cardiac Anomalies Only	6	11%
	ASD	3	
	VSD	1	
	PDA	2	
	Pectus	1	
Post Operative Complications		8	15%
	Wound Infection	2	1.90%
	Pneumonia	6	11%
	PO Morbidity	7	
	PO Mortality	1	
Pathology	Abnormalities Of Bronchial Cartilage	7	13%
	Polyalveolar Lobe	1	2%

of the pleural cavity and insertion of 2 chest drains with debridement of the wound and re-closure. Post operative pneumonia reported in 6 cases where three cases managed by antibiotics and 3 cases subjected to mechanical ventilation because of respiratory failure, 2 cases weaned from mechanical ventilation after 4 and 7 days respectively and the third case died after 10 days of mechanical ventilation from sepsis. Pathological examination performed routinely for all resected specimens, where abnormalities of bronchial cartilage tissue were reported in seven cases, polyalveolar lobe in one case, no specific etiological factors could be detected in the remaining cases. All the previously mentioned results were summarized in Table 1. In all cases chest drain removed after (1–3) days post-operatively except for one case which developed severe wound infection and encysted empyema, the drains removed 8 days after the re-insertion. Except for complicated cases, all patients discharged from hospital after 4–6 days postoperatively. Late postoperative outcome was satisfactory, as all survivors were followed up for one year at outpatient clinic and all of them were free of symptoms and within normal X-ray findings.

4. Discussion

CLE is a rare cause of respiratory distress in neonates and early infantile life [23]. CLE commonly seen in males more than females [24]. In the present study; thirty nine were males and fourteen were females. These findings are consistent with the literature [23]. Lobe affection is usually unilateral with the left upper lobe is the most commonly affected (40–50%) followed by the middle lobe (25–30%) then the right upper lobe (20%). Lower lobe affection is extremely rare (2–5%) [25]. Lower lobe affection was reported by some authors [18,26]. Bilateral lobar involvement is very rare [27] however it has been reported in some series [28]. Staged management is usually practiced in case of bilateral involvement [28]. In the current series; there was no lower lobe affection or multiple lobar diseases. The left upper lobe was involved in 66% of cases, followed by right upper lobe in 19% cases and the right middle lobe in 15% cases. Some series reported nearly the same order of anatomical disease distribution as in our study [18,29,30]. The difference of anatomical distribution of CLE between different reports may be due to the relatively small number of patients in these series or due to racial cause, the exact cause of CLE is difficult to determine. The etiology seems to be multifactorial. Progressive lobar hyperinflation is the end result of a number of intra-uterine deviations of broncho-pulmonary development which may lead to a change in the anatomy of airways, alveoli or both [31]. Surgical treatment of CLE with concomitant congenital heart disease have been reported to be simultaneous or staged but in any way the combination of congenital heart disease and CLE increases the risk and is associated with poor outcome especially if the congenital heart disease is of complex type [32]. In our series, there was only one case of left upper lobe emphysema managed at the same time with PDA ligation and the case gone well. No definite cause is found in more than 50% of cases, in the remaining patients, the most distinct abnormality is congenital bronchial cartilage defect of the affected lobe. Other reported causes of bronchial obstruction include intrinsic web, mucous folds, mucous plugging [30]. Bronchial obstruction from outside by vascular abnormalities or intra-thoracic masses is a rare causes [24]. Polyalveolar lobe is another theory which attributes CLE to increased number of alveoli in each acinus [33]. Findings of histopathological examination of the resected lobes in our series showed abnormalities of bronchial cartilage in 13% of cases and polyalveolar lobe in 2%, and no definite cause was detected in 85% of cases. Nearly the same results reported by others [12,30]. Diagnosis of CLE is challenging, the main presenting symptoms of CLE related to respiratory distress are characterized by dyspnea, tachypnea, wheezing,

cough, and cyanosis [26]. In severe cases, the thoracic wall over the involved side is more prominent, respiratory excursion is limited and breath sounds are diminished over the affected lobe [9,18]. Older infants present mainly with repeated chest infection [10]. The above-mentioned symptoms and clinical findings were also detected in our patients. In our series, signs of respiratory distress were the commonest presenting signs.

Once diagnosis of CLE is suspected, CXR is the initial step to evaluate the patient in the current series as well others [12,30]. In our series, a confirmatory pre-operative CT chest was done for 75% of cases. In the remaining 25%, we depended only on CXR because the operation was urgent in 7 cases and in another 6 patients the CT machine in our hospital was out of service. In another Egyptian series published in 2001, the authors stated that they depended on only CXR in 66% of cases and pre-operative CT was done only for 33% of cases [30]. This may be attributed to the cost and unavailability of CT during the era of study period which started in 1975 in comparison to the relatively widespread use and facility of CT during the study period of our series. None of the patients in our study was diagnosed before birth, however, some authors have reported that CLE can be diagnosed prenatally by ultrasound [10,34]. The differential diagnosis includes congenital pulmonary airway malformation (CPAM), bronchopulmonary sequestration, bronchogenic cyst, and congenital diaphragmatic hernia, pneumothorax, pulmonary hypoplasia, pneumatocele, endobronchial obstruction and huge mediastinal bronchogenic cyst [35–38]. CXR of the chest in CLE can sometimes confuse physicians and to be misdiagnosed as tension pneumothorax which lead to unnecessary intercostal tube (ICT) insertion. Many authors reported surgical treatment of cases of CLE with history of inadvertently inserted ICT for false diagnosis of CLE as tension pneumothorax [17,36,39–41]. In our series, we have operated three cases with history of false diagnosis as tension pneumothorax and ICT insertion was done at emergency department of peripheral hospitals before referral to our tertiary hospital. The role of bronchoscopy in the diagnosis of CLE is limited, it is not a primary screening test, it was used only in patients over six months of age to exclude foreign body inhalation or obstruction by thick secretion plugs and avoid inappropriate surgery, in accordance with the algorithm proposed by Karnak et al. [12]. We performed bronchoscopy for seven cases pre-operatively; five rigid and two fiber-optic in infants older than six months to rule out to foreign body inhalation and all were negative. The same findings also reported by others [12,18]. This also in agreement with Dr kadry and his colleagues who believe that bronchoscopy is not indicated in the classical cases of hyperlucent lung except in older infants in whom inhalation of foreign body is suspected as a cause of bronchial obstruction [30]. Routine Preoperative echocardiographic examination is mandatory as cardiac anomalies are associated in up to 20% of cases of CLE [42]. In our series, we had reported 6 cases (11%) associated with cardiac anomalies. In addition to cardiac anomalies, a large number of abnormalities have been described in association with CLE, such as pectus excavatum, anterior mediastinal defect, diaphragmatic hernia, hiatal hernia, chondroectodermal dysplasia, chondro-dystrophy, aplastic kidney, cleft palate, and pyloric stenosis, cystinosis, and an omphalocele [1,9,10]. In our series, pectus excavatum was reported in one case.

The classically accepted treatment for CLE is Lobectomy [10,30]. As regard conservative treatment of CLE, it is a great matter of controversy as many reports stated that conservative therapy should be avoided [1,18,19]. Some authors stated that when CLE patients managed conservatively, 50% of them died from progressive respiratory distress, and 75% of survivors has persistent emphysema [30]. On the other hand, an increasing number of reports showed that many patients with CLE can be safely managed conservatively [10,20,21,43,44]. Actually, there is no struggling between surgical treatment and conservative treatment of CLE. We and others believe that the choice of treatment is based on severity of respiratory distress. Clearly, severe respiratory distress is an indication for surgical treatment in infants with CLE, while, conservative treatment is justified in patients with no or mild symptoms. However, careful selection and strict follow up are needed since deterioration may occur necessitating surgical intervention [6,10,39]. Other reports discuss the benefits of segmental lung resection for CLE instead of anatomical lobectomy but there is a great risk of recurrence of symptoms [45]. We are in agreement with protocol of karank [12] and protocol of kadry [30] in management of CLE; they stated that: in neonates and infants with severe respiratory distress, emergency thoracotomy is life saving, for less acute cases, elective thoracotomy is performed. In our series, all cases of CLE were treated surgically by lobectomy. Left upper lobectomy was the most frequent procedure (66%) followed by right upper lobectomy (19%) and then middle lobectomy (15%).

An important anesthetic aspect during induction is to avoid over inflation and high positive pressure as this could compromise hemodynamic and ventilator states [46]. It is considered the most critical time which is the time between induction of anesthesia and the opening of the chest and delivery of the over-distended spongy emphysematous lobe outside the chest [19]. Raghavendra and his colleagues reported the use of thoracic epidural analgesia and spontaneous breathing till the chest is opened [47]. High frequency jet ventilation was tried by Goto et al. [48]. Our anesthesia team was very helpful as regard this issue, anesthesia started after the sedated patient was draped and the surgeon was standby for thoracotomy. Very gentle manual ventilation started with the use of thoracic epidural analgesia as shot single dose to be effective until chest is opened and emphysematous lobe delivered outside the chest then anesthesia completed as usual and ventilator started. The difference between our protocol in giving epidural analgesia and others is to give a single shot dose without insertion of catheter.

Post operative pneumonia was the most common complication in our series and others [24,49]. Three patients out of six who developed pneumonia culminating into respiratory failure necessitating mechanical ventilation, one of them died. Mortality rate in our series was 1.9%. Review of published reports revealed a wide range of mortality from 0% up to 21% [1,10,12,18,24,26,32,49] and this wide range of percentage of mortality is due to many factors like the number of patient in each series [30], the percentage of association of congenital anomalies and either operated at the same time or not, the presence of pre-operative pneumonia, pre-operative mechanical ventilation [49] and the time of reporting the series as

actually every new year we found advancement in the equipment, operation rooms, ventilation and anesthesia which actually increase the success rate and decrease the mortality. Our results are acceptable and encouraging with 98.1% survival rate which matches with the results of series within the range of patient number and similar patient and hospital criteria. We are in agreement with Elmenshaway [49] as he stated that history of pre-operative mechanical ventilation increase the risk of poor outcome, the one patients who died postoperatively in our series was operated on urgent basis while he was on mechanical ventilation pre-operatively because of respiratory failure and chest infection not responding to medical treatment.

5. Conclusions

Surgical treatment of CLE by means of lobectomy is a safe procedure and the active co-operation between the departments of cardiothoracic surgery, pediatric, radiology and anesthesia directly affect the success rate of diagnosis and management. Early diagnosis and management is a golden key for a favorable outcome.

Conflicts of interest

The authors whose names are listed immediately below certify that they have NO affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, knowledge or beliefs) in the subject matter or materials discussed in this manuscript.

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