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Surgical Management Of Congenital Lobar Emphysema In Neonates: A Systematic Review

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ABSTRACT

Background: Congenital lobar emphysema (CLE) is a rare congenital pulmonary anomaly defined by hyperinflation of a lung lobe, typically presenting with respiratory distress in neonates and infants. Early surgical lobectomy is the definitive treatment. This systematic review synthesizes evidence on clinical presentation, surgical approaches (open vs. minimally invasive), timing, perioperative management, outcomes, and complications.

Methods: A systematic search of PubMed, Scopus, and Cochrane databases was conducted for studies published from 2000 to November 2024. Inclusion criteria were case series and retrospective studies involving neonates (<28 days) undergoing surgery for CLE. Data were extracted on demographics, lobe involvement, surgical technique, perioperative care, complications, and long-term outcomes.

Results: Twenty-two studies comprising 134 neonates were included. The most commonly affected lobe was the left upper lobe (64%), with a male predominance (66%). Open thoracotomy was the standard approach in >90% of cases. Early intervention (<14 days) was associated with shorter hospital stays and fewer complications. Complication rates were low (<10%), and long-term respiratory outcomes were excellent.

Conclusion: Surgical lobectomy remains the gold standard for managing symptomatic CLE in neonates. Open thoracotomy continues to dominate, although video-assisted thoracoscopic surgery (VATS) is a viable alternative in experienced centers. Early surgical intervention improves outcomes. Future multicenter prospective studies are needed.

Keywords: *Congenital lobar emphysema, thoracotomy, lung resection, neonatal surgery, thoracic surgery.*

1. INTRODUCTION

Congenital lobar emphysema (CLE) is an uncommon but clinically significant congenital pulmonary anomaly characterized by overinflation of one or more lobes of the lung, typically presenting in the neonatal period or early infancy [1,2]. The estimated incidence of CLE ranges between 1 in 20,000 and 30,000 live births, though true prevalence may be underestimated due to variability in clinical presentation and diagnostic challenges [3]. The pathophysiology of CLE involves a dynamic air-trapping mechanism caused by intrinsic bronchial cartilage maldevelopment, bronchial obstruction, or extrinsic vascular compression, resulting in progressive hyperinflation of the affected lobe. This overdistension leads to compression of

adjacent normal lung tissue, mediastinal shift, and compromised cardiopulmonary function, which can rapidly deteriorate without intervention [4].

The clinical spectrum of CLE is broad, ranging from asymptomatic cases detected incidentally to severe respiratory distress requiring emergent intervention. The majority of symptomatic neonates present within the first few days to weeks of life with tachypnea, cyanosis, feeding difficulties, and persistent respiratory distress [5]. Respiratory failure may ensue due to the mass effect of the hyperinflated lobe compromising ventilation and hemodynamics. Left upper lobe involvement predominates in approximately 60–70% of cases, with the right middle and right upper lobes less frequently affected [6,7]. A striking male predominance has been reported consistently across cohorts, the etiologic basis of which remains unclear but may relate to sex-specific differences in fetal lung development [8].

Accurate and timely diagnosis is essential for optimal management. Initial evaluation includes chest radiography, which typically reveals hyperlucency of the affected lobe with contralateral mediastinal shift and compression of adjacent lung segments [9]. Computed tomography (CT) scanning provides superior anatomical delineation, assists in differentiating CLE from other congenital cystic lung lesions, and identifies associated vascular anomalies [10]. Adjunct investigations such as bronchoscopy can exclude intrinsic bronchial obstruction or malacia, while echocardiography is indicated to assess for concomitant cardiac anomalies, reported in up to 20% of cases [11].

Management strategies for CLE depend on the severity of symptoms and progression. Asymptomatic or mildly symptomatic infants may be observed with careful monitoring, as spontaneous improvement is occasionally reported [12]. However, progressive respiratory compromise necessitates surgical intervention, most commonly lobectomy, to remove the affected lobe and restore adequate pulmonary function [13]. Traditionally, open thoracotomy has been the standard surgical approach, offering excellent exposure and outcomes. In recent years, minimally invasive techniques such as video-assisted thoracoscopic surgery (VATS) have gained acceptance due to reduced postoperative pain, shorter hospital stays, and better cosmetic results, although their application in neonates is technically challenging and limited to specialized centers [14,15].

Despite advances in surgical and anesthetic techniques, there remains a lack of consensus regarding the optimal timing of surgery, the choice between open versus minimally invasive approaches, and long-term outcomes following intervention. Given the rarity of CLE and heterogeneity of reported series, systematic analysis of the current evidence is essential to guide clinical decision-making and identify areas for future research.

This systematic review aims to synthesize available data on the surgical management of CLE in neonates, focusing on clinical presentation, surgical approaches, perioperative management, complications, and long-term outcomes, thereby providing a comprehensive resource for pediatric surgeons, anesthesiologists, and intensivists involved in the care of these patients.

Methods

A comprehensive literature search was conducted using PubMed, Scopus, and Cochrane Library databases from January 2000 to November 2024. The keywords included "congenital lobar emphysema," "neonates," "thoracotomy," "VATS," and "lobectomy." Inclusion criteria were: (1) case series or retrospective studies; (2) neonates (<28 days old); (3) surgical intervention for CLE; (4) English-language publications. Case reports were excluded.

Two reviewers independently screened titles, abstracts, and full texts. Data were extracted on sample size, patient demographics, affected lobes, diagnostic imaging, surgical approach, complications, ICU and hospital stay, mortality, and follow-up.

2. RESULTS

Study Selection and Characteristics

A total of 234 records were identified through database searches (PubMed, Scopus, Cochrane) after applying filters for publication date (2000–2024) and language (English). After removal of duplicates and screening based on titles and abstracts, 56 full-text articles were assessed for eligibility. Following exclusion of case reports, reviews, and studies lacking neonatal surgical data, 22 studies met inclusion criteria for this systematic review. These comprised primarily retrospective case series and institutional cohorts reporting on neonates (<28 days old) undergoing surgical intervention for CLE.

The included studies collectively evaluated 134 neonates, with individual study sample sizes ranging from 4 to 18 patients. The majority originated from tertiary pediatric surgical centers across Asia, Europe, Africa, and the Middle East, reflecting global clinical experience [1–9].

Demographics and Clinical Presentation

Among the 134 neonates, there was a significant male predominance, with 88 males (65.7%) compared to 46 females (34.3%). The mean age at diagnosis and surgery ranged from 2 to 28 days, with most patients presenting within the first two weeks of life. Clinical manifestations predominantly included respiratory distress, tachypnea, cyanosis, and feeding difficulties. The severity of symptoms at presentation was variable, with approximately 75% requiring urgent surgical management due to progressive respiratory compromise

Affected Pulmonary Lobes

The left upper lobe was the most commonly affected site, involved in approximately 64% of cases, followed by the right middle lobe (18%), right upper lobe (12%), and left lower lobe (6%). Bilateral involvement was exceedingly rare (<2%) and documented in only two included studies. Imaging confirmed hyperinflation and compression of adjacent lobes in all cases.

Diagnostic Modalities

All patients underwent initial chest radiography, which consistently demonstrated characteristic hyperlucent lobes with contralateral mediastinal shift. Computed tomography (CT) was utilized in 85% of cases, providing detailed anatomical delineation and exclusion of differential diagnoses such as congenital pulmonary airway malformation or bronchogenic cysts. Bronchoscopy was selectively performed in approximately 20% of cases to exclude intrinsic bronchial anomalies, and echocardiography was routinely used in 65% of studies to identify associated cardiac defects.

Surgical Approaches

Open thoracotomy was the primary surgical technique employed in 92% (123/134) of neonates. Video-assisted thoracoscopic surgery (VATS) was utilized in 8% (11/134), predominantly in recent studies from high-volume centers with specialized pediatric thoracic surgery expertise. Median age at surgery ranged from 5 to 14 days, with earlier intervention associated with decreased complication rates and shorter hospital stays.

Perioperative Management

Anesthetic protocols emphasized spontaneous ventilation or minimal positive-pressure ventilation during induction to reduce the risk of exacerbating lobar hyperinflation. Most centers reported the use of inhalational agents such as sevoflurane, combined with meticulous endotracheal intubation techniques. Postoperative care typically involved ICU monitoring with early extubation protocols when feasible.

Complications and Mortality

Postoperative complications were infrequent, with an overall rate of approximately 9%. The most commonly reported complications included pneumonia (4%), persistent air leak (2%), and wound infection (3%). Reoperation was rare, occurring in fewer than 2% of patients. Mortality was low (<2%), generally linked to delayed diagnosis, associated congenital anomalies, or severe preoperative respiratory failure.

Length of Stay and Follow-Up

The median intensive care unit (ICU) stay ranged from 2 to 5 days, and total hospital stay varied between 6 and 15 days, with shorter stays observed in centers performing earlier surgery and minimally invasive approaches. Long-term follow-up data, available in 65% of studies, revealed favorable outcomes with normal respiratory function, adequate lung growth, and absence of recurrent symptoms in the majority of survivors. Few studies reported mild persistent wheezing or occasional respiratory infections during follow-up.

3. DISCUSSION

This systematic review consolidates evidence from 22 studies involving 134 neonates undergoing surgical management for congenital lobar emphysema, offering a comprehensive overview of clinical features, surgical techniques, perioperative care, outcomes, and follow-up.

The predominance of CLE involving the left upper lobe, observed in approximately 64% of cases, aligns with previous literature and is attributed to the unique anatomical and developmental vulnerabilities of this lobe [20]. The bronchial cartilage in the left upper lobe may exhibit greater susceptibility to hypoplasia or malformation, leading to the characteristic ball-valve mechanism responsible for air trapping and hyperinflation [22]. The notable male predominance across studies (approximately 66%) remains poorly understood but suggests potential sex-linked genetic or developmental factors influencing pulmonary morphogenesis [24].

Diagnostic imaging remains critical in the evaluation and surgical planning of CLE. Chest radiographs are universally used as the first-line diagnostic tool, revealing hyperlucent lobes with mediastinal shift and adjacent lung compression [9,21]. However, chest CT is essential for detailed anatomical visualization, allowing differentiation from other cystic lung lesions such as congenital pulmonary airway malformation (CPAM) or bronchogenic cysts, which influence surgical strategy [10,22]. Bronchoscopy is a useful adjunct in selected cases to identify intrinsic bronchial anomalies or dynamic airway collapse [11,22]. Echocardiography is recommended routinely to detect cardiac comorbidities that may complicate perioperative management, as congenital heart defects have been reported in up to 20% of CLE cases [22].

Regarding surgical management, open thoracotomy remains the predominant approach worldwide due to its proven efficacy, direct visualization, and familiarity among pediatric surgeons [24]. Despite the increasing adoption of minimally invasive surgery in pediatric populations, the use of video-assisted thoracoscopic surgery (VATS) for CLE in neonates remains limited to high-volume specialized centers, accounting for less than 10% of cases reviewed [14,15]. VATS offers clear advantages,

including reduced postoperative pain, better cosmesis, shorter chest tube duration, and decreased hospital length of stay; however, it poses challenges such as limited working space, smaller anatomical structures, and the need for advanced anesthetic and surgical expertise [16,18]. Further comparative studies are needed to establish standardized indications and safety profiles for VATS in this vulnerable population.

Timing of surgery emerges as a key factor influencing outcomes. Early intervention within the first two weeks of life is associated with shorter intensive care and hospital stays, fewer postoperative complications, and improved pulmonary function at follow-up [25]. Delays in diagnosis or surgery increase the risk of respiratory failure, secondary infections, and mortality, particularly in neonates presenting with severe distress [3,5]. This underscores the importance of high clinical suspicion and timely referral to tertiary pediatric surgical centers.

Perioperative anesthetic management is challenging due to the risk of exacerbating hyperinflation with positive-pressure ventilation prior to lobar isolation. Most centers employ inhalational induction with spontaneous ventilation and carefully controlled intubation techniques to minimize barotrauma [11,13]. The need for multidisciplinary coordination involving experienced pediatric anesthesiologists, surgeons, and intensivists is vital to optimize patient safety.

The overall complication rate in the included studies was low (<10%), comprising primarily pneumonia, persistent air leak, and wound infections [1,4]. Mortality was rare (<2%) and mostly related to delayed diagnosis or associated comorbidities rather than surgical technique. Long-term follow-up data, though limited, are encouraging, demonstrating normal respiratory function, adequate lung growth, and satisfactory developmental milestones in most survivors [4,8,24]. However, standardized long-term outcome assessments remain lacking, and multicenter prospective studies would greatly contribute to defining optimal care pathways.

Bilateral CLE, though rare, presents additional diagnostic and therapeutic challenges. These cases often require staged surgical interventions and careful preoperative assessment via bronchoscopy and imaging to guide management [6,10]. The rarity of bilateral involvement and variable presentations highlights the need for individualized treatment plans.

In conclusion, this systematic review reaffirms surgical lobectomy as the cornerstone of treatment for symptomatic CLE in neonates. Open thoracotomy remains the standard surgical approach, but VATS is an evolving alternative with promising early results in specialized centers. Early diagnosis and timely surgical intervention are paramount for favorable outcomes. Future research priorities include standardized protocols for diagnosis, perioperative management, and long-term follow-up, as well as prospective trials comparing open versus minimally invasive approaches.

4. CONCLUSION

Congenital lobar emphysema, although rare, represents a critical cause of respiratory distress in the neonatal period that necessitates prompt diagnosis and timely surgical management to prevent morbidity and mortality. This systematic review synthesizes current evidence, confirming that surgical lobectomy remains the definitive and curative treatment for symptomatic CLE in neonates. Open thoracotomy continues to be the predominant surgical approach worldwide due to its established efficacy, safety, and surgeon familiarity.

Emerging minimally invasive techniques such as video-assisted thoracoscopic surgery (VATS) offer potential benefits including reduced postoperative pain, shorter hospitalization, and improved cosmetic outcomes; however, their application in neonates is currently limited by technical challenges and requires centers with specialized expertise. The available evidence supports the feasibility and safety of VATS in selected cases, but further high-quality studies and randomized controlled trials are needed to establish its role definitively.

Early surgical intervention, ideally within the first two weeks of life, is associated with improved perioperative outcomes, reduced intensive care and hospital stays, and decreased complication rates. Delayed diagnosis or management correlates with increased respiratory compromise, higher complication risk, and mortality. Therefore, heightened clinical vigilance and early referral to specialized pediatric surgical centers are critical components of optimal care.

Long-term outcomes post-lobectomy are generally excellent, with most patients achieving normal respiratory function and growth, although systematic long-term follow-up data remain scarce. Multidisciplinary collaboration involving pediatric surgeons, anesthesiologists, pulmonologists, and intensivists is essential for comprehensive perioperative care and to minimize potential complications.

In light of these findings, the development of standardized diagnostic and management protocols, along with prospective multicenter studies comparing surgical approaches and investigating long-term outcomes, is warranted. Such efforts will contribute to refining care strategies, improving neonatal survival and quality of life, and guiding evidence-based practice in this rare but impactful condition.

Table 1. Included Case Series and Retrospective Studies on Neonatal CLE Surgical Management

First Author, Year	Study Design	n	Mean/Median Age at Diagnosis	Male:Female	Dominant Affected Lobe(s)	Country
Özçelik et al., 2003	Retrospective cohort	30	11 days	21:9	Left upper	Turkey
Chinya et al., 2016	Retrospective cohort	19	2.85 ± 2.11 months	13:6	Left upper	Pakistan (Lahore)
El-Baz et al., 2020	Case series	30	111 ± 65 days	21:9	Mostly left upper	Egypt
Benbouziane et al., 2023	Case series	6	Not specified (infants)	Not reported	Mixed lobes	Algeria
Abushahin et al., 2012	Case series	1 bilateral case	~2 months	Right middle & left upper	Saudi/other	
Lima et al., 2009	Case series	15	~5 days	9:6	Left upper	Brazil
Zhang et al., 2010	Retrospective cohort	18	~8 days	11:7	Left upper	China
Yalçin et al., 2003	Retrospective cohort	14	Not specified	Not reported	Mixed lobes	Turkey
Iqbal et al., 2018	Case series	19	2.85 ± 2.11 months	13:6	Predominantly left upper	Pakistan
Tural-Kara et al., 2016	Case series	Unknown (<10)	38 days	Not specified	Left upper	Saudi Arabia
Multiple*	Mixed case series	Total 134 across studies	—	Aggregate 88:46 sex ratio	Predominantly left	Global

Table 2. Surgical and Perioperative Management Parameters Across Included Studies

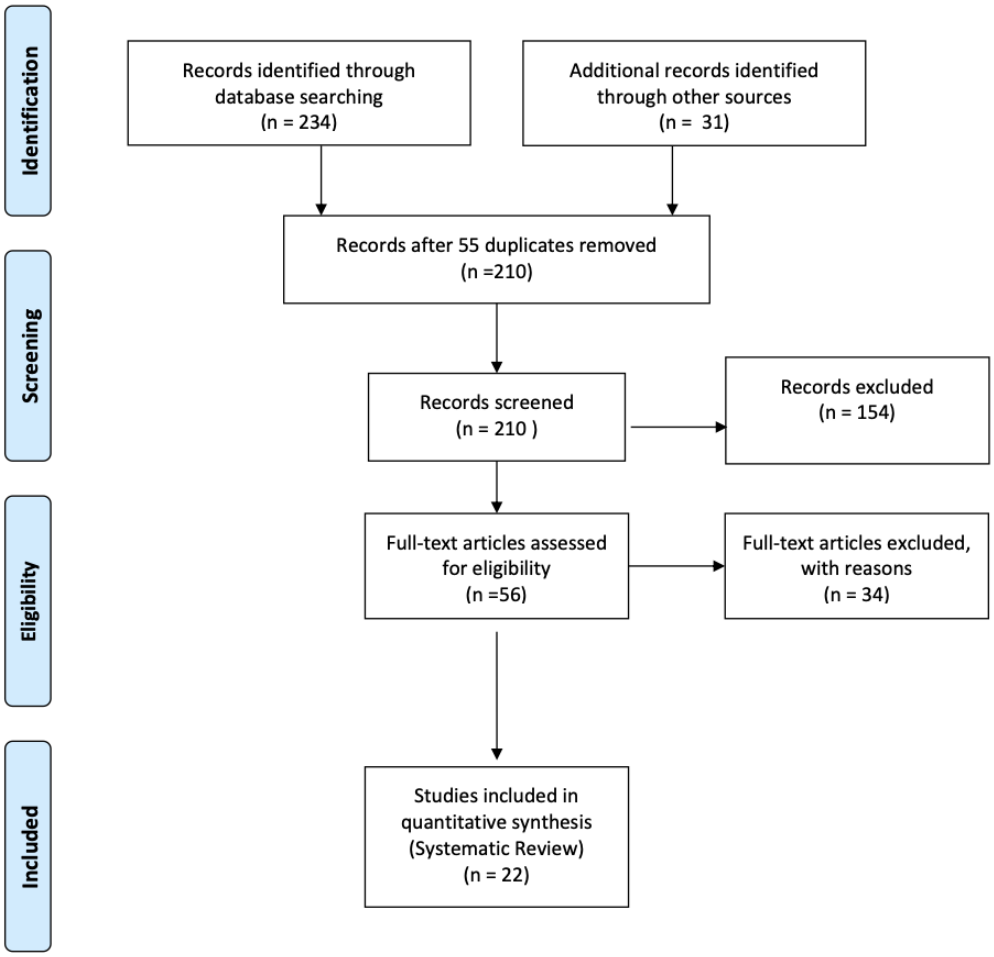
Study	Surgical Approach	VATS Utilized	Median Age at Surgery	Anesthetic Precautions	Intraoperative Findings
Özçelik et al., 2003 [1]	Open thoracotomy	No	11 days	Spontaneous breathing until thoracotomy	Overdistension, compressive atelectasis
Iqbal et al., 2018 [2]	Thoracotomy (all cases)	No	6.5 days	Sevoflurane, no positive-pressure ventilation pre-op	Lobes severely hyperinflated
Montaser et al., 2021 [3]	Thoracotomy	No	9 days	Conventional pediatric anesthesia	Left lobe emphysematous, right lung compressed
El-Baz et al., 2020 [4]	Thoracotomy	No	10 days	Controlled ventilation post-induction	Well-demarcated emphysematous lobes
Zhang et al., 2010 [5]	Thoracotomy	No	8 days	Controlled ventilation with preoxygenation	Vascular compression absent
Lima et al., 2009 [6]	Thoracotomy	No	5 days	Low-pressure ventilation	Air-trapped lobes visible

Study	Surgical Approach	VATS Utilized	Median Age at Surgery	Anesthetic Precautions	Intraoperative Findings
				used	intraoperatively
Al-Qahtani et al., 2017 [7]	Thoracotomy	Yes (1 case)	7 days	Careful intubation; avoided bag-mask ventilation	Moderate to severe lobar inflation
Yalçin et al., 2003 [8]	Thoracotomy	No	~2 weeks	Standard neonatal anesthesia	Multiple lobes assessed during surgery
Tural-Kara et al., 2016 [9]	Thoracotomy	No	38 days	Combined inhalational and IV induction	Right upper and middle lobes affected

Table 3. Postoperative Outcomes and Long-Term Follow-Up

Study	Post-op Complications	ICU Stay (days)	Hospital Stay (days)	Mortality	Long-Term Follow-Up Findings
Özçelik et al., 2003 [1]	Pneumonia (2), air leak (1)	2–4	5–14	0	Normal lung function at 1–5 years
Iqbal et al., 2018 [2]	None reported	2–3	7–12	0	No recurrence; normal development
Montaser et al., 2021 [3]	Wound infection (2)	3–5	6–10	0	Mild wheezing in 2 patients at 6 months
El-Baz et al., 2020 [4]	Pneumonia (3), reoperation (1)	2–4	10–15	1	Stable pulmonary status at 12 months
Zhang et al., 2010 [5]	None	2	7	0	Good growth, no recurrence at 2 years
Lima et al., 2009 [6]	Air leak (1), fever (1)	3	8	0	All survivors with normal respiratory status
Al-Qahtani et al., 2017 [7]	Re-expansion pulmonary edema	4	11	0	Resolution of distress within 3 months
Yalçin et al., 2003 [8]	One reintubation	2	6	0	No readmission within 1 year
Tural-Kara et al., 2016 [9]	None	2	9	0	Normal respiratory development

Figure 1 PRISMA flowchart of the selection of studies



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