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Review Article

Sublobar Pulmonary Resection in Children With Congenital Lung Abnormalities: A Systematic Review

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A B S T R A C T

Background: Lobectomy is currently the advised resection for symptomatic congenital lung abnormalities (CLA). Sublobar surgery is suggested as an alternative that enables to conserve healthy lung parenchyma. This systematic review aims to explore the outcomes of sublobar surgery in CLA patients as well as the corresponding surgical terminology and techniques.
Methods: A systematic literature search was performed in adherence to PRISMA-P guidelines. The target population consists of children undergoing sublobar pulmonary resection for CLA. All studies were independently assessed by two reviewers, and evaluated by a third reviewer in case of disagreement.
Results: The literature search yielded 901 studies of which 18 studies were included, comprising 1167 cases. The median chest tube insertion duration was 3.6 days (range 2.0–6.9 days), the median hospital admission was 4.9 days (range 2.0–14.5 days), and residual disease was diagnosed in 2% – leading to reoperation in 70%. The median incidence of postoperative complications was 15% (range 0–67%). Follow-up imaging was standard-of-care in 2/3 of studies. Due to the absence of standardised terminology, operative details and specification of resection type did not typically relate between studies.
Conclusions: Sublobar resection of CLA lesions could be a viable alternative to lobectomy in certain cases, with the advantage of conserving healthy lung parenchyma. Peri- and postoperative complications are comparable with those reported for conventional lobectomy. The incidence of residual disease following sublobar surgery appears to be lower than commonly stated. To improve comparability between studies, we recommend reporting perioperative characteristics in a structured format.
Level of evidence: Level IV.

1. Introduction

Congenital lung abnormalities (CLA) is an umbrella term for pulmonary anomalies that include, among others, cystic pulmonary airway malformation (CPAM), bronchopulmonary sequestration (BPS), bronchopulmonary cysts (BC), and congenital lobar overinflation (CLO) [1–3]. Between 3 and 20% of CLA patients present with postnatal symptoms, which primarily include respiratory distress, recurrent lung infection or persistent cough [4–6]. In these cases, surgical resection is indicated [7–9]. However, in case a child is asymptomatic after birth, there is still ongoing debate between those in favour of conservative wait-and-see management and those that prefer elective surgery [9]. In this debate, arguments posed in favour of conservative management include the risks associated with major surgery in young children and the possibility of overtreatment, considering that the majority of initially asymptomatic children seem to remain symptom-free during childhood [8,10,11]. On the other hand, common arguments in favour of surgical resection are the possible prevention of symptom development, the increased complication risk of surgery once symptoms have presented, and the risk of malignant transformation in CLA lesions [5,8,12,13].

In any case, if surgery is chosen, a lobectomy is currently the advised resection in children with CLA [8,13]. Sublobar surgery – also referred to as wedge resection, sublobar resection or segmentectomy – is proposed as an alternative for lobectomy, with the

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Abbreviations

AL	Persistent air leakage
AT	Atelectasis
BC	Bronchogenic cyst
BF	Bronchopulmonary fistula
BPS	Bronchopulmonary sequestration
CLA	Congenital lung abnormalities
CLO	Congenital lobar overinflation
CPAM	Congenital pulmonary airway malformation
CT	Computed tomography
HE	haemorrhage
KRAS	Kirsten rat sarcoma virus
PA	Pneumonia
PE	Pleural effusion
PI	Pulmonary infection
PC	Persistent cough
P-CT	Pulmonary perfusion CT
PN	Persistent pneumothorax
PRISMA-P	Preferred reporting items for systematic reviews and meta-analysis protocols
PROSPERO	Prospective registry of systematic reviews
RD	Respiratory insufficiency/distress
RL	remnant lesion
SE	Subcutaneous emphysema
VATS	Video-assisted thoracoscopic surgery
WD	Wound dehiscence
WI	Wound infection

potential to preserve healthy lung parenchyma [5,6,14–16]. In adult literature, sublobar surgery has been accepted as an alternative treatment for benign disease, carcinoid tumours, or pulmonary metastases [17,18]. In addition, sublobar surgery has been proven to result in superior survival and pulmonary function when compared to lobectomy in selected cases of early-stage lung cancer [19,20]. Despite this, and the promising prospect of saving healthy lung parenchyma, sublobar surgery has not been generally adopted as treatment of choice in the CLA population, even though CLA lesions are generally considered a benign disease [11]. An often-mentioned argument against sublobar surgery is the risk of residual disease, the rate of which mentioned in literature is as high as 15%, with unclear clinical consequences so far [8,21,22].

Sublobar operations can be divided in anatomical (i.e., segmentectomy) and non-anatomical (i.e., excision, wedge resection) resections, depending on whether the resection adheres to segmental anatomic borders, or that margins are chosen based on lesion boundaries, optical or radiological cues. An anatomical sublobar resection is technically demanding, because the lung parenchyma needs to be dissected further towards the periphery to identify the segment level anatomy, ligate segmental vessels and bronchi, and define the parenchymal borders of the segment(s). The segmental anatomy is especially demanding in paediatric CLA patients due to the smaller scale of segmental structures and a large number of anatomical variations [16]. In adult patients undergoing segmentectomy for early-stage lung cancer, segmentectomy has been found associated with a safety profile comparable to that of lobectomy, albeit with an increased risk for prolonged post-operative air-leakage [23]. However, it is unknown if the increased difficulties associated with segmentectomy in children lead to higher complication rates in the paediatric CLA population.

At present, inter-study comparability is hindered by inconsequent use of terminology [14,24,25]. Therefore, we firstly aim to provide a systematic overview of the peri- and postoperative outcome of sublobar surgery in paediatric patients with a CLA. Secondly, recommendations or guidelines regarding the terminology, documentation of the resection, and outcome for sublobar resections in CLA cases do not exist, to our knowledge. Hence, we provide an overview of the terminology reported and the surgical techniques used to provide recommendations for a uniform documentation.

2. Methods

2.1. Study protocol

This review was performed according to ‘Preferred Reporting Items for Systematic Reviews and Meta-analysis Protocols (PRISMA-P) systematic review guidelines [26,27]. The completed checklist is shown in the supplementary materials. This review included the following five key phases: (1) identifying the research question, (2) identifying relevant studies, (3) study selection, (4) charting the data, and (5) collating, summarizing, and reporting the results. The study protocol was registered online in the PROSPERO prospective registry of systematic reviews (Registration number CRD42022345823) [28].

3. Research aim

The primary aim of this systematic review was to provide a comprehensive overview of the perioperative and postoperative outcomes in paediatric patients with CLA following sublobar surgery. Secondarily, surgical details and terminology as described in literature were collected.

3.1. Data sources, search strategy and study management

The predefined search was performed among the electronic libraries Embase, Medline, Cochrane and Web of Science in collaboration with the librarians of the Erasmus University Medical Center in April 2021 and updated in November 2022. No date limits were placed on the database search. All studies initially retrieved from the search were imported into the web-based systematic research software Rayyan for title and abstract screening, after which all included studies were imported into the bibliographic manager Endnote 20 [29].

3.2. Eligibility criteria

Eligible studies were those with a cohort, case–control, or randomized controlled trial design, written in English, and available in full-text format. Review studies and studies including fewer than three paediatric patients were excluded. The population consists of paediatric patients (<18 years) with a sublobar pulmonary resection for a CLA.

3.3. Title, abstract and full-text screening

Two reviewers (CMK and JJW) independently assessed the eligible studies for compliance with the inclusion and exclusion criteria by title and abstract. Subsequently, CMK and MR performed full text analysis of the selected studies to select the studies for final inclusion. Both for title and abstract and full text assessment, discrepancies were discussed until consensus was reached and a third reviewer (JMS) was consulted if necessary.

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3.4. Data summary and synthesis

The following extracted data were charted: general study characteristics (title, authors, country, year of publication, type of article), patient characteristics (number, age at surgery, sex, symptoms), CLA characteristics (type of CLA, diagnostic methods, timing of diagnosis, asymptomatic/symptomatic), treatment characteristics (type of resection, surgical access, conversion, surgery duration, parenchymal division, determination of resection borders, parenchyma sealing method) and outcome characteristics (complications, duration of chest tube insertion, length of hospital admission, follow-up imaging and duration of follow up, residual disease, re-operation).

4. Results

4.1. Study selection and characteristics

The flowchart depicted in Fig. 1 summarizes the study selection procedure. The complete online search thread can be found in the supplementary materials. The initial search (01–2020) yielded 770 studies, to which 131 studies were added following a search update (11–2022), leading to a total of 901. Based on title and abstract screening, 86 remained for full text assessment, leading to the eventual inclusion of 18 studies [14,15,21,30–44]. The study characteristics of the 18 included studies are listed in Table 1.

4.2. Patient characteristics

The included studies encompassed 1167 cases which consisted of 446 CPAM, 378 BPS, 25 CLE, 1 BC, 45 Hybrid lesions, 180 cystic lesions, and 92 unspecified lesions. The median age at time of surgery was 15.2 months (range 0.03–204 months). The median proportion of symptomatic patients was 32% (range 4–100%). The symptoms consisted of persistent cough, respiratory distress, and/or pulmonary infection. Most studies (72%) used chest computed tomography (CT) to confirm the diagnosis, in some cases (11%) preceded by a chest X-ray. Additional details can be found in Table 1.

4.3. Operative details

Operations were mostly reported as segmentectomy. Other used terminology included lung-sparing resection, sublobar resection, wedge resection, fractionated lung resection, lung-sparing segmental resection, atypical resection or anatomical lesion resection. Surgical access was obtained through minimally invasive methods (video-assisted thoracoscopic surgery – VATS) in seven studies, through thoracotomy in three studies, through both VATS and thoracotomy in six studies, while two studies did not specify the method of access. Ten studies addressed conversion rate (from thoracoscopic to thoracotomic resection), with eight studies noting no conversions, and two studies with a conversion rate of 33% an 35%, respectively. Median surgery duration ranged from 56 to 140 min, and parenchymal dissection was performed with either Ligasure, Endostaplers, Endo GIA, Ultrasonic scalpel, or regular Electrocautery or sutures.

Thirteen studies described intra-operative steps in detail, one of which described two different methods. In seven of them, individual ligation of bronchial branches and vessels was initially performed, followed by dissection of the segmental parenchyma of interest. These procedures were reported as segmentectomy or lung-sparing segmental resection. Two other studies noted transection along non-anatomical segmental lines following visible, palpable, or radiographically detected lines, and documented these procedures as either segmentectomy or lung-sparing resection, the

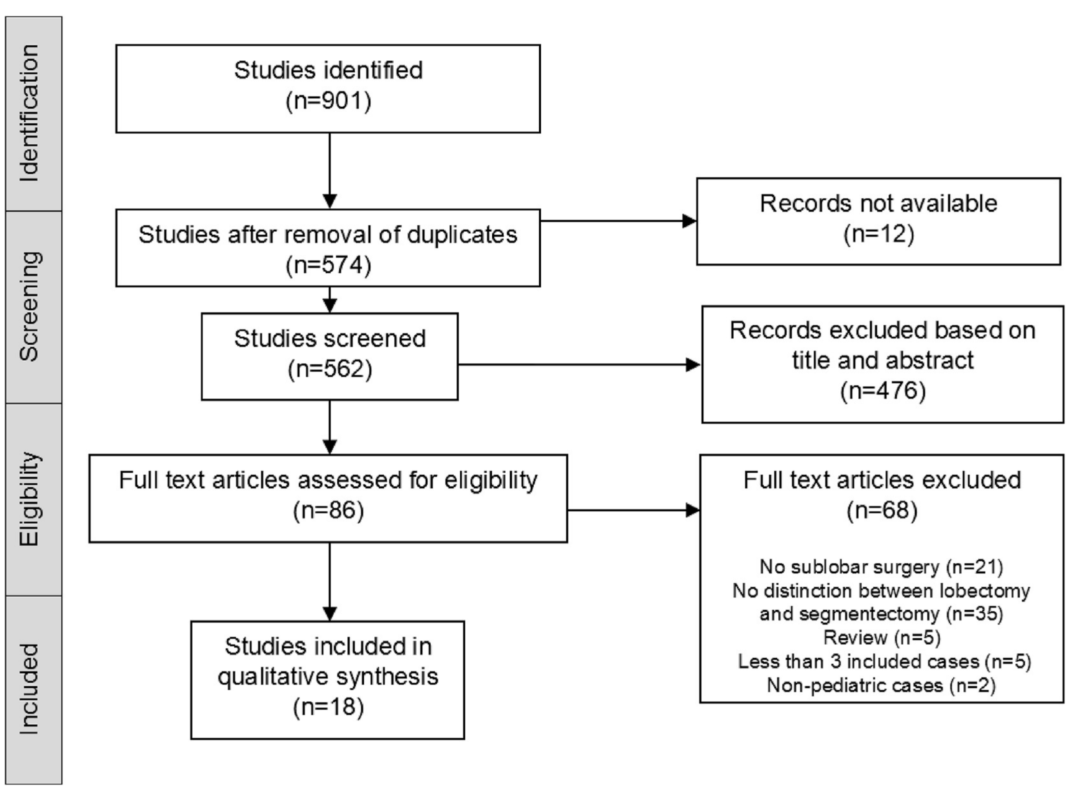

Fig. 1. Flowchart of study selection.

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Table 1
Patient characteristics.

Author	N	Year	Country	Study type	Diagnoses (N)	Gender (N)	Age at surgery Median in months (range), ±SD	Symptomatic (%)	Symptoms	Diagnosis	Timing of diagnosis
Bagrodia [14]	19	2014	USA	Retrospective cohort	n.a.	M 10, F 9	7.0 (2–180)	32	RD, PI, PC	chest CT	prenatal 58%
Cheng [40]	206	2022	China	Retrospective cohort	CPAM 159, BPS 45, CLE 2	M 95, F 115	6 (3–156)	n.a.	n.a.	n.a.	n.a.
Erginel [30]	9	2015	Turkey	Retrospective cohort	CPAM 9	M 4, F 5	74.4 (1–204)	n.a.	n.a.	chest CT	n.a.
Fascetti-Leon [31]	54	2013	Italy	Retrospective cohort	CPAM 23, Hybrid 17, BPS 10, BC 1, Other 3	M 26, F 28	8.6 (0.5–168)	18	RD, PI	chest CT	prenatal 92%
Galazka [44]	11	2020	Poland	Retrospective cohort	BPS 4, CPAM 1, BPS 2, Hybrid 4	M 4, F 7	8 (4–21)	n.a.	n.a.	n.a.	prenatal 75%
Guo [42]	16	2022	China	Retrospective cohort	CPAM 16	M 12, F 4	7.2 (3.8–70.0)	44	RD, PI	chest-CT	prenatal 94%
Huang [43]	19	2021	China	Retrospective cohort	n.a.	M 12, F 7	4.4 ± 0.8	n.a.	n.a.	n.a.	n.a.
Ito [32]	4	2019	Japan	Retrospective cohort	CPAM 4	M 3, F 1	0.3 (0.1–39)	100	RD, PI	chest CT	prenatal 75%
Johnson [33]	15	2011	USA	Retrospective cohort	n.a.	n.a.	n.a. (6–204)	40	RD, PI	chest X-ray, chest CT, MRI	prenatal 80%
Keidar [21]	3	2001	Israel	Retrospective cohort	CPAM 3	n.a.	n.a.	0	n.a.	chest CT	prenatal 100%
Kim [34]	12	2008	South Korea	Retrospective cohort	n.a.	M 7, F 5	51.6 (n.a.)	67	PI	chest CT	prenatal 13%
Krivchenya [15]	22	2013	Ukraine	Retrospective cohort	CLE 22	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.
Lee [35]	20	2017	South Korea	Retrospective cohort	n.a.	n.a.	3.9 (0.03–8)	n.a.	RD	chest CT	prenatal 100%
Lilly [36]	5	1976	USA	Retrospective cohort	CPAM 4, CLE 1	M 4, F 1	2,7 (0,2–7,4)	100	RD	chest X-ray, Bronchography, P-CT	postnatal 100%
Lima [37]	167	2019	Italy	Retrospective cohort	CPAM 167	M 96, F 71	12.3 (0.4–204.0)	30	RD, PI	chest CT	prenatal 76%
Polites [38]	486	2016	USA	Retrospective cohort	Cystic 180, BPS 282, Hybrid 24	M 268, F 218	21 ± 50	4	PI	n.a.	n.a.
Tarrado [39]	4	2015	Spain	Retrospective cohort	BPS 4	M 2, F 2	10.5 (9.0–180.0)	25	Hemoptysis	chest CT	prenatal 75%
Yuan [41]	95	2022	China	Retrospective cohort	CPAM 60, BPS 35	M 64, F 31	10 (5–36)	n.a.	n.a.	chest-CT	n.a.

N.a. = not available, M = male, F = female, CLE = congenital lobar emfysema – more recently known as CLO = congenital lobar overinflation, CPAM = congenital pulmonary airway malformation, BC = bronchogenic cyst, BPS = bronchopulmonary sequestration, cyst, Hybrid = hybrid lesion, RD = respiratory distress, PI = pulmonary infection, PC = persistent cough, chest CT = computed tomography, chest X-ray = plain chest radiography, MRI = magnetic resonance imaging, P-CT = Pulmonary perfusion CT.

latter further categorised as segmentectomy and atypical resection. Two studies retained the intra-segmental vein as the internal border, dissected towards the external border from there, and finally ligated the bronchial branches and vessels. These procedures were referred to as modified wedge resection or anatomical lesion resection. Two other studies noted segment dissection starting from the parenchymal periphery, and referred to the procedures as segmentectomies. Lastly, one study described the use of methylene blue for infusion in the feeding artery of a bronchopulmonary sequestration, subsequently marking the borders with monopolar cautery, and documented this procedure as a segmentectomy.

In addition, three of these 13 studies described the observation of ischemic changes after clipping of the vascular structures to help identify segmental borders. Two other studies described the same process, only aided by ventilation distinction after clipping of the segmental bronchus. Parenchymal sealing methods varied, and included various sealing devices but also absorbable running sutures, sealing of the suture lines with local visceral pleura and enveloping with polyglycolic acid patches and fibrin glue. Full operative details are displayed in Table 2 and the supplementary materials. The extracted surgical information from the included studies was used to design a structured surgical report for paediatric lung resections (Fig. 2). Additionally, a schematic representation was designed, depicting the separate types of resections with a distinction between anatomical and non-anatomical resections (Fig. 3).

4.4. Outcome

Sixteen studies described the complication rate; among those the median complication rate was 15% (range 0–67) and 131 out of the 1136 patients included in these studies developed complications, leading to a total complication rate of 12%. These complications primarily included persistent pneumothorax (17), persistent air leakage (11), pneumonia (6), respiratory distress (4), haemorrhage (3). The median chest tube insertion duration was 3.6 days (range 2.0–6.9) and were primarily left inserted due to air leak. The median hospital admission duration was 4.9 days (range 2.0–14.5). Only three studies discussed the need for blood transfusions which was necessary in 0–5% of these cases [31,33,37]. Fifteen studies reported on residual disease following surgery and reported this in 0–67% of cases per study (median 0%), and in 2% of all these cases (N = 10). Re-operation was undertaken in 70% of residual disease cases, but it was unclear if these cases presented with symptoms.

Table 2
Operative details.

Author	Terminology	Surgical access	Conversion	Surgery duration	Parenchymal division	DeterminSiation of resection borders	Anatomical or non-anatomical resection	Parenchyma sealing
			N (%)	Mean minutes (range), ±SD				
Bagrodia	Segmentectomy	VATS 79%,	0	n.a.	LigaSure, Endostapler	n.a.	n.a.	LigaSure, Endostapler
Cheng	Segmentectomy	VATS 100%,	0	56 (45–112)	Ligasure, Ultrasonic scalpel	Ischemic changes after clipping of vascularisation + ventilation boundary after clipping of bronchus	Anatomical by means of ischemic + ventilation differentiation	Ligasure, Ultrasonic scalpel
Erginel	Segmentectomy	VATS 33%,	n.a.	n.a.	LigaSure, Endo GIA	n.a.	n.a.	LigaSure, Endo GIA
Fascetti-Leon	Lung-sparing resection (Segmentectomy and Atypical resection)	VATS 48%,	18 (33%)	122 (65–255)	LigaSure, Bipolar cautery, Endostapler	Atypical resection: non-anatomical visible/palpable/radioloical borders. Segmentectomy: anatomical boundaries	Non-anatomical lines	Absorbable running sutures
Galazka	Excision, Lung-sparing surgery, Segmentectomy	VATS 100%,	0	140 (75–200)	BiClamp	Ischemic changes after clipping of vascularisation/optical delineation based on border of hyperaemic parenchyma	Anatomical by means of ventilation differentiation or optical determination of hyperaemic border	n.a.
Guo	Modified thoracoscopic wedge resection	VATS 100%,	0	74 (50–110)	Ligasure, Ultrasonic scalpel	Identification of intra-segmental vein	Anatomical by means of identification of intra-segmental vein	Ligasure, Ultrasonic scalpel
Huang	Segmentectomy	VATS 100%,	0	87 ± 134	Ligasure, Ultrasonic scalpel	Ventilation boundary after clipping of bronchus	Anatomical by means of ventilation differentiation	Ligasure, Ultrasonic scalpel
Ito	Segmentectomy, Wedge resection, Fractionated lung resection	VATS 25%	n.a.	78 (67–107)	n.a.	n.a.	n.a.	n.a.
Johnson	Segmentectomy	VATS 100%,	0	n.a. (90–240)	LigaSure, Endostapler	Visual and radiographic borders	Non-anatomical lines	Ligasure, Endostapler
Keidar	Segmentectomy	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.
Kim	Segmentectomy, Wedge resection	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.
Krivchenya	Lung-sparing segmental resection	Thoracotomy 100%	n.a.	n.a.	Endostapler, sutures	n.a.	n.a.	Sealing suture lines with peeled off local visceral pleura
Lee	Segmentectomy	Thoracotomy 100%	n.a.	127 ± 38	Electrocautery, LigaSure	CT evaluation + ventilation after dissection of segmental bronchi	Anatomical by means of ventilation differentiation	Eveloping with polyglycolic acid patches and fibrin glue
Lilly	Segmentectomy	Thoracotomy 100%	n.a.	n.a.	n.a.	Identification of intersegmental vein	Anatomical by means of identification of intersegmental vein	n.a.
Lima	Segmentectomy, Atypical resection	VATS 40%	29 (35%)	109 ± 40	n.a.	n.a.	n.a.	n.a.
Polites	Segmental resection, Sub-lobar resection	VATS 48%	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.
Tarrado	Segmentectomy	VATS 100%	0	120 (55–240)	Endostapler	Coloring of lesion with methylen blue and marking with monopolar cautery	Anatomical by means of methylene blue	Endostapler
Yuan	Thoracoscopic anatomical lesion resection	VATS 100%	0	63,2 ± 15,2	Ligasure, Ultrasonic scalpel	Emphysema-like boundary, circulation boundary, pulmonary lobular units, or tortuous increased blood vessels	Anatomical by means of optical differentiation,	Ligasure, Ultrasonic scalpel

N.a. = not available, VATS: Video-assisted thoracoscopic surgery.

Structured surgical report Congenital Lung Abnormalities

Patient characteristics:

Patient age (months):

Surgical circumstances: ☐ Elective ☐ Emergency

Thoracic epidural: ☐ yes / ☐ no

Single-lung ventilation: ☐ yes / ☐ no

Antibiotics: ☐ yes / ☐ no

Symptoms: ☐ yes / ☐ no

If yes, type of symptoms:

☐ Respiratory insufficiency ☐ Failure to thrive

☐ Recurrent infection ☐ Cardiac overload

☐ Other: ....

Lesion characteristics:

Side: ☐ left ☐ right ☐ bilateral

Lobe:

RUL	RML	RLL	LUL	LLL

Size: .... x .... x .... mm

Diagnosis (suspected):

☐ CPAM ☐ Bronchopulmonary Sequestration

☐ Bronchogenic Cyst ☐ Congenital Lobar Overinflation

☐ Other:

Surgical procedure

Access

☐ Thoracoscopy

☐ Thoracoscopy with intra-operative conversion

☐ Thoracotomy

Please note indication: ....

Operation time: ..... min

Intra-operative complications:

☐ Blood loss, ... ml

☐ Air leakage

☐ Other: ....

Anatomical resection:

☐ Lobectomy, please specify:

RUL	RML	RLL	LUL	LLL

☐ Segmentectomy, please specify:

R	L	1	2	3	4	5	6	7	8	9	10

Mode of identification anatomical boundaries:

☐ Ventilation differentiation ☐ Contrast agent (e.g., Methylene blue, ICG) ☐ Other: ....

☐ Vascular differentiation ☐ Imaging (pre-operative / during surgery)

------------- OR -----------------

Non-anatomical resection:

☐ Wedge resection ☐ Other: ....

☐ Cystectomy

Post-operative management

Post-operative recovery

☐ Intensive Care ☐ High Care ☐ Conventional ward

Thoracic drain:

☐ No / ☐ Yes, 1 / ☐ Yes, 2 / ☐ Yes, 3

☐ Suction .... cmH2O / ☐ Water seal

Drain management: ....

Post-operative imaging:

☐ Imaging modality: ☐ Chest X-ray / ☐ Other: ....

☐ Timing after surgery: ....

Remarks:

Remarks:

Conclusion:

Fig. 2. Structured surgical report Congenital Lung Abnormalities.

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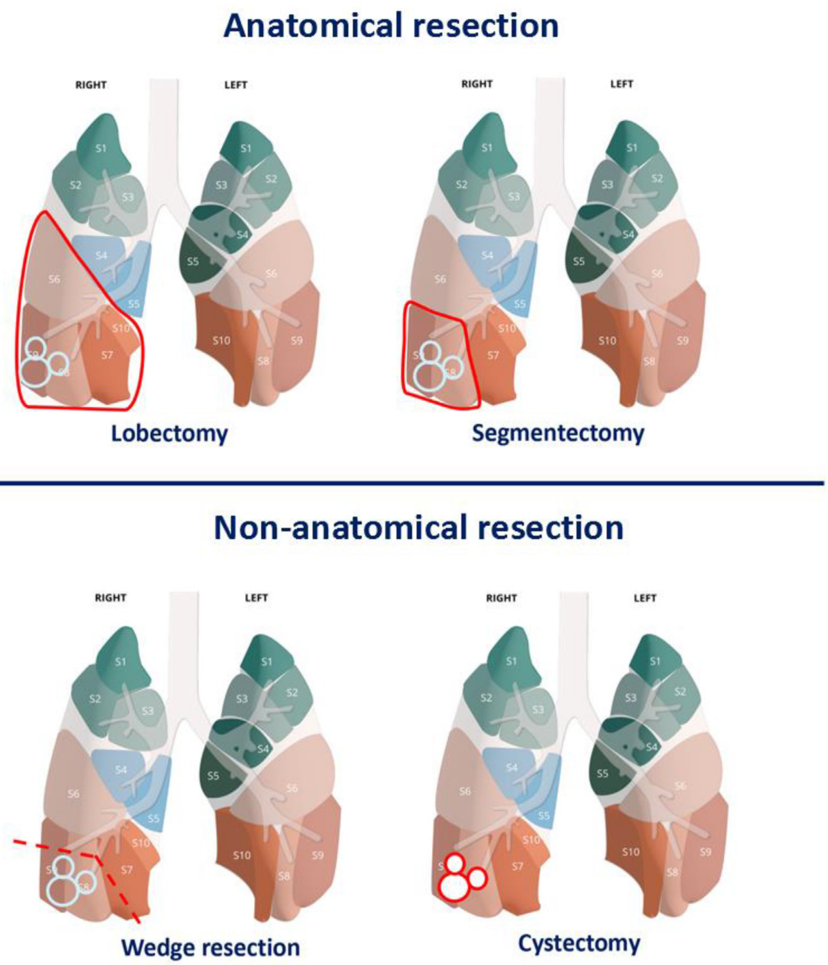

Fig. 3. Schematic depiction of types of pulmonary resection, distinguishing between anatomical resections – Lobectomy and Segmentectomy -, and non-anatomical resections – Wedge resection and Cystectomy. A continuous red line indicates the dissection border, while a dotted red line indicates the positioning of a stapler or sealer device for dissection. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

The median follow-up duration was 23 months (range 10–65), which included standard-of-care imaging in 12 out of 18 studies, primarily by means of chest-CT imaging. No cases of malignancy were encountered on histological examination, neither was any mortality observed. Full details of the outcome characteristics are presented in Table 3.

5. Discussion

This review provides a comprehensive overview of sublobar resections in children with a congenital pulmonary abnormality, a surgical procedure that has the potential to save lung parenchyma and improve long-term lung function [6,14]. The included studies comprise 1167 cases and report a combined complication rate of 15%. This rate falls within the range of 10%–25% from large-scale reports on paediatric lobectomies [45,46]. Likewise, the duration of chest tube insertion and length of hospital stay – 3.6 days and 4.9 days respectively – were also in line with findings from available studies on lobectomies [45,47,48]. This review found the frequency of residual disease to be 2%, considerably lower than the 15% reported before [8]. One possible explanation for this discrepancy is that only one study overlapped between these two reviews, presumably due to the difference in systematic search strategy motivated by dissimilar research questions.

Most children whose residual disease was confirmed (70%) underwent re-operation by means of lobectomy. In those studies, follow-up imaging was standard of care, but insufficient information was available on the presence of symptoms in cases with residual disease. These findings do not support the implementation of standard of care follow-up imaging, for which no evidence is available and caution is advised considering the exposure to harmful radiation. In addition, possible oncogenic parameters such as Kirsten rat sarcoma virus (KRAS) mutations or Dicer 1 were not described [49]. Neither was the presence of mucinous proliferations documented, which is a morphological trait detected in the majority of adenocarcinoma in situ among CPAM cases [50]. It remains thus unclear to what extent residual disease has clinical relevance, and in which specific cases re-operation is indeed required [51–53].

The risk of residual disease is an often-posed argument against sublobar resection in children. Most likely, this risk can be diminished by optimal preoperative imaging and planning – for example with help of 3D virtual reality software – thereby increasing the accuracy of adhering to the anatomic segmental borders in the subsequent resection [54]. This argument is supported by the recent advances in the treatment of early-stage lung cancer in adults, for which sublobar surgery has been accepted as an alternative treatment [17–20,55]. However, CLA lesions differ from solid

Table 3
Outcome characteristics.

Author	Complications	Complication details	Chest tube duration	Blood transfusion	Length of hospital admittance	Follow up imaging	Residual disease	Re-operation	Follow up
	N (%)		Mean days (range), ±SD	N (%)	Median days (range), ±SD		N (%)	N (%)	Median months (range), ±SD
Bagrodia	4 (21%)	2 PN, 1 PE, 1x WI	2 (0–12)	n.a.	2 (1–12)	chest X-ray	0	0	10 (0–97)
Cheng	6 (3%)	5 AL, 1 AT	1 (1–14)	n.a.	3 (3–14)	chest CT	0	0	12 (3–75)
Erginel	n.a.	n.a.	3.6 (2–7)	n.a.	4.8 (3–8)	Not standard	0	0	n.a.
Fascetti-Leon	6 (11%)	3 HE, 3 PN	5.5 (1–27)	3 (6%)	8 (7–45)	chest X-ray, chest CT in case of symptoms	1 (2%)	1 (2%)	65 (12–115)
Galazka	0	n.a.	1.5 (0–4)	n.a.	4.9 (3–8)	n.a.	n.a.	n.a.	n.a.
Guo	2 (13%)	2 SE	3 (2–4)	n.a.	6 (4–8)	chest CT	0	0	10 (3–18)
Huang	3 (16%)	3 PN	4 ± 0.9	n.a.	5.1 ± 0.8	chest CT	0	0	24
Ito	1 (25%)	1 RL	3.5 (3–4)	n.a.	14.5 (9–16)	n.a.	1 (25%)	0	n.a.
Johnson	5 (33%)	3 PN, 1 AL, 1 BF	3.7 (0–21)	0	4.2 (1–21)	chest CT	1 (7%)	1 (7%)	18 (7–43)
Keidar	2 (67%)	2 RL	n.a.	n.a.	4 (4–6)	chest CT	2 (67%)	1 (33%)	n.a.
Kim	1 (9%)	1 AL	5.3 ± 3,7	n.a.	7.8 ± 4,0	chest CT in case of symptoms (67%)	1 (8%)	0	64 ± 44
Krivchenya	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	0	0	n.a.
Lee	4 (20%)	3 AL, 1 WD	6.9 (3–22)	n.a.	10.8 (8–24)	chest CT	1 (5%)	1 (5%)	23 ± 12.9
Lilly	1 (25%)	1 AL	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.	n.a.
Lima	15 (9%)	6 PN, 4 PA, 4 RD	5.8 ± 4.5	8 (5%)	9 (7–45)	chest CT	3 (2%)	3 (2%)	65 (12–115)
Polites	79 (16%)	n.a.	n.a.	n.a.	2.5 and 4*	n.a.	n.a.	n.a.	n.a.
Tarrado	0	n.a.	2.5 (2–3)	n.a.	3 (2–5)	chest CT	0	0	24 (9–38)
Yuan	2 (2%)	2 PA	1.3 ± (1–2)	n.a.	3.3 ± (3–5)	chest CT	0	0	12

N.a. = not available, chest X-ray = plain chest radiography, chest CT = thoracic computed tomography, PN = persistent pneumothorax, PE = pleural effusion, WI = wound infection, AL = persistent air leakage, AT = atelectasis, HE = haemorrhage, SE = subcutaneous emphysema, RL = remnant lesion, BF = bronchopulmonary fistula, WD = wound dehiscence, PA = pneumonia, RD = respiratory distress.

tumours in structural composition as well as growth pattern, including distant adjacent lesions that could be missed during sublobar surgery [56]. Nevertheless, sublobar surgery for this generally considered benign disease should at least be considered.

Undeniably, anatomical sublobar surgery in children comes with additional challenges. Apart from increased technical difficulty due to the small scale of essential structures, the increased complexity of pre-operative radiological evaluation and the small working area, surgeons should be aware that children with CLA often exhibit anatomical variations [16,54]. However, the potential of preserving healthy lung parenchyma may very well benefit the child's long-term exercise capacity and lung function – which are important outcome measures for the management of CLA [9,20]. Only one study is available that compares postoperative lung function between sublobar surgery and lobectomy cases, and reports on improved lung function in the former, although several studies have reported on normal long term lung function in children that underwent lobectomy at early age [43,57,58]. More research into the functional outcomes after paediatric lung surgery is thus needed. Among the studies included in this review, the performed pulmonary resections were documented in a quite diverse manner. From the provided operative details, it was apparent that terminology was not used in a consistent manner, as provided surgical details could well be similar, even though the assigned terminology differed or vice versa. The included articles described several methods for the identification of intersegmental borders, such as ventilation or ischemic differentiation after clamping of the segmental bronchus or arteries, dissection along the intra- or intersegmental vein, or intravenous administration of the dye methylene blue. However, five studies did not provide these operative details, on account of which it was not clear whether the procedures were either anatomical or non-anatomical resections. A structured method for reporting surgical details could help to describe procedures in a standardised way, facilitate comparison of future studies, and bring to light possible outcome differences between anatomical and non-anatomical resections. For that reason, we designed a structured surgical report, as shown in Fig. 2.

Despite the comprehensive literature search that served as a basis for this systematic review, several limitations need to be addressed. First, the included studies had quite high extents of missing data, especially concerning follow-up characteristics. The implementation of standardised follow-up protocols could possibly diminish these missing data [9,59]. Several inclusion criteria also led to limitations. For example, the inclusion of all types of CLA is a limitation, because the different types may require different management strategies and may differ in expected outcome. Still, to increase the number of included surgical procedures for this review, we chose to include all studies in which sublobar resections were undertaken in children with a CLA. Moreover, only retrospective studies in the English language were included which could possibly introduce a bias in the results.

As for future studies reporting on surgical procedures in paediatric CLA patients, we suggest to use a uniform terminology to describe surgical procedures, as for example provided in Fig. 2. In addition, we plea for distinguishing between anatomical resections – comprising of lobectomy and segmentectomy – and non-anatomical resections – comprising of wedge resection, cystectomy, and so on. A schematic representation of the distinction between these resection types is shown in Fig. 3.

Clear and uniform reporting of surgical details will improve comparability between studies, especially necessary for rare diseases such as CLA. Moreover, the role of contrast agents such as indocyanine green or methylene blue combined with virtual reality planned segmentectomy – as already described in adult cases –

could help improving anatomical resection accuracy in paediatric cases [60–62]. Further exploration into the feasibility of these new innovative methods for children is definitely indicated [63].

In conclusion, sublobar resection of CLA lesions could be a safe and viable alternative to lobectomy in certain eligible cases, with the advantage of conserving healthy lung parenchyma. The incidences of peri- and postoperative complications are comparable with those encountered with conventional lobectomy, and from this review it appears that the incidence of residual disease following sublobar surgery is lower than formerly reported. In order to improve comparability between studies, we recommend to report pre-, per, and postoperative characteristics in a structured format.

Previous communication

Not applicable.

Conflict of interest

The authors have no relevant financial or non-financial interests to disclose.

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Appendix A. Supplementary data

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