Original Research Article

Short term outcome of lung resection in children-a preliminary report from India

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Received: 06-02-2021 / Revised: 15-03-2021 / Accepted: 08-04-2021

Abstract

Background : With advancement in technologylung resection surgeries are commonly being done in children for various congenital and acquired reasons refractory to medical management. Consequently, it becomes imperativeto assess their outcomes, short and long term, and association, if any with resection technique. In the present study, researchers intend to study short term outcome of lung resection in such children, who underwent resection at their institute, a tertiary care centrefrom North India. **Methodology**:A retrospective review of the patients who underwent lung resection for a benign condition (CLL and acquired lung disease), from 2000-2011 was done. Children aged more than 4 years at the time of evaluation and at least one year post surgery were included in the study. Children with associated co-morbidities or had undergone non anatomical lung resection or resection performed for empyema thoracis or a mass lesion like teratoma were excluded. Participants were subjected to clinical, radiological and spirometric evaluation.**Result :**Twenty patients at a mean follow up of 3.41 years and mean age of 7.55 years (± SD 3.69) were evaluated for final analysis. Most common indication for surgery was congenital lung lesions (CLL) (65%). Pulmonary function was assessed only in 45% (n = 9) of the patients and it showed mild restriction. Winging of scapula most common musculoskeletal problem seen in children who undergone muscle cutting incision for thoracotomy. Most patients were clinically well. Only two patients, had recurrent cough and required readmissions for respiratory infections. Most patients (90%) showed good somatic growth. All cases had shown compensatory growth of the remaining lung tissue, irrespective of the age at surgery and the indication for surgery.**Conclusion:**We conclude that lung resection surgery is well tolerated by these children.Child attained good somatic growth and remaining lung tissue grew well to compensate, irrespective of the age and the indication for surgery. Musculoskeletal abnormalities are more following muscle cutting incision.

Keywords : Lung resection in children, Pulmonary function test, congenital lung lesion, winging of scapula.

Introduction

Although lung resection procedures are comparatively rare [1]in children, with advancement in science these operations are now being performed more frequently in neonates and children for various congenital and acquired lung lesions. [2]Most common indication is congenital lung lesions (CLL), while acquired lung diseases, including infective and neoplastic lesion is next important indication. Literature on the outcomes of these cases in terms of pulmonary function test, exercise tolerance, effect on growth and development of the child is scanty, particularly from India. [1, 3, 4, 5]The existing literature gives a controversial picture and often represent the western setup, but the indications and presentation differs widely among the developed and the developing countries.

We, therefore, aimed to study lung functions and clinical outcomes in children who underwent lung resection surgery, using spirometry for pulmonary function tests and compared these values with standard nomograms. We have also assessed the clinical and radiological outcomes in these cases.

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Materials and methods

After obtaining institutional ethical committee approval, a retrospective review of the patients who underwent lung resection for a benign condition (CLL and acquired lung disease), from 2000-2011 was done. Children aged more than 4 years at the time of evaluation and at least one year postresection were included in the study. Children with associated co-morbidities (congenital heart disease/ chest wall anomalies/ thoracic vertebral anomalies) or had undergone non anatomical lung resection or resection performed for empyema thoracis or a mass lesion like teratoma were excluded. Thirty one patient fulfilled selection criteria. Among these only 24 could be contacted through available contact details, among whom twenty patients responded and were included in the study. These patients were subjected to clinical, radiological and spirometric evaluation.

Data was collected for demographic details, clinical presentation at the time of surgery, surgical details and post op follow up details till the time of inclusion in the study. Evaluation included clinical examination, chest X ray and pulmonary function test. Clinical examination included anthropometry, general examination with special reference to musculoskeletal examination- winging of scapula, kypho-scoliosis, respiratory system examination, and mean oxygen saturation at room air. Chest X ray- (postero-anterior and lateral view) was done for position of trachea and mediastinum, herniation and hyperinflation of contralateral lung, bilateral lung

parenchyma, rib cage, pleural fibrosis and spinal curvature.Scoliosis was defined as a lateral spinal curvature with a Cobb angle of 10° or more and if scoliosis present, cobb`s angle was measured manually. [6] Pulmonary Function Test [PFT] was performed in paediatric pulmonology laboratory by using Schiller TM spirometer. [7, 8] Following parameters were recorded in spirometry: Peak expiratory flow (PEF), Forced vital capacity (FVC), Forced expiratory volume in 1 second (FEV1), FEV1/FVC. These parameters were compared with established standard reference values and expressed as percentage predicted. Analysis of data obtained from spirometry was used to classify type of abnormalities as either obstructive, restrictive, combined obstructive and restrictive pattern. The degree of pulmonary dysfunction was assigned as mild, moderate and severe as shown in below mentioned Figure 1.

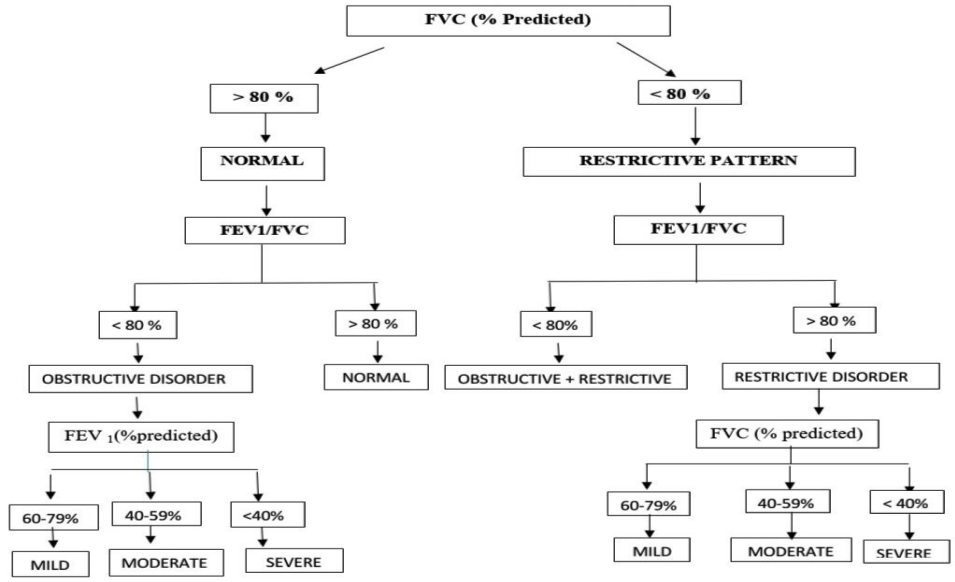

Fig 1: Degree of pulmonary dysfunction

Children with obstructive pattern were further assessed for reversibility of airflow obstruction after inhalational bronchodilator (salbutamol, 400 mcg). If FEV1 improved by 15% or more from its base line (pre bronchodilator) value then obstruction was labelled as reversible.The data so obtained was correlated with the surgical details of the patient and analysed using SPSS version 20. Inference was derived with regard to clinical and functional outcome in children following lung resection.

Results

In twenty patients, the assessment for clinical, radiological and pulmonary function outcome was carried out at a mean follow up of 3.41 years. Mean age of the patients at the time of evaluation was 7.55 years (± SD 3.69). Most common indication for surgery was congenital lung lesions (CLL) (65%), among whom CCAM [Congenital cystic adenomatoid malformation] (40%) was the most common lesion followed by CLE [Congenital lobar emphysema] (20%). Bronchiectasis was the most common indication (15%) among acquired lung disease.Pulmonary function was assessed only in 45% (n = 9) of the patients as rest of the patients could not cooperate for spirometryand it showed mild restriction (Table 1.)

Table 1: Demographic details, clinical assessment and pulmonary functions of the patients

Patient no.	Age (years	Age at surgery	Indication/ Surgery/Incision	Clinical/Musculoskeletal Observations	PFT
1	14/M	11.5	FB induced chronically destroyed Lt lung/ Pneumonectomy/MC	Winging of scapula	Reversible obstruction
2	4/F	0.7	Rt lower lobe CCAM /Lobectomy/MS	-	-
3	4/F	0.3	Lt lower lobe CCAM/Lobectomy/MS	Winging of scapula	-
4	5.5/M	0.5	Lt upper lobe CLE/ Lobectomy/MS	-	-
5	4/M	0.05	Lt upper lobe CLE/Lobectomy/MS	-	-
6	10/F	7.5	Complicated hydatid cyst Lt lower lobe/Lobectomy/MC	Winging of scapula	Mild restriction
7	9/M	1.8	Lt UL Necrotising pneumonia, pneumatocoel/Lobectomy/MC	-	Moderate restriction
8	5/M	0.21	Lt lower lobe CCAM/Lobectomy/MS	-	-
9	11/M	8.5	Bronchopulmonary forgut malformation/RU lobectomy/MC	-	Severe restriction with partially reversible obstruction

10	10/F	7	Recurrent LCH Lt main bronchus/Pneumonectomy/MC	Winging of scapula	Mild restriction
11	6/M	0.5	RLL CCAM/Lobectomy/MC	-	-
12	14/F	12.25	Infected CCAM Rt Lower lobe/Lobectomy/MC	-	Mild restriction
13	4.2/M	0.17	CCAM Rt upper and middle lobe/ RUL & RML Lobectomy/ MS	-	-
14	7/F	5.5	CCAM Lt upper and lingular lobe/Lobectomy/MS	Winging of scapula/ Lt anterolateral depression of chest wall	-
15	4/F	0.33	CLE Rt Upper and Middle lobe / Lobectomy/ MC	-	-
16	4/M	0.75	CCAM Rt upper and middle lobe/Lobectomy/ MS	-	-
17	6/M	5	Post tubercular Lt atelectobronchiectasis/pneumonectomy/MC	Winging of scapula/ Pectus carinatum	Reversible obstruction
18	4.2/F	0.008	CLE Lt Upper Lobe/ lobectomy /MC	-	-
19	12/M	9.6	Bronchiectasis Lt lower lobe/ lobectomy/MC	-	Mild restriction
20	13/M	11	Bronchiectasis Rt lung/Pneumonectomy/ MC	Winging of scapula	Mild restriction

(M=Male, F=Female, Rt = Right, Lt = Left, MC = Muscle cutting, MS = Muscle sparing, CCAM=Congenital cystic adenomatoid malformation, CLE=Congenital lobar emphysema, RLL= Right lower lobectomy, RUL = Right upper lobectomy, RML = Right middle lobectomy, LCH = Langerhans' cell histiocytosis)

All our patients had posterolateral thoracotomy with muscle cutting incision in 60% and muscle sparing in 40% cases. Six patients required postoperative ventilator support. Significant post-operative complications were seen in two cases. None of our patient had wound infection. Mean hospital stay was 13.95 days. It was longer for those having muscle cutting incision as compared to muscle sparing group incision. The difference could not reach statistical significance. On clinical evaluation, most patients were clinically well. Only two patients, had recurrent cough and required readmissions for respiratory infections. Most patients (90%) showed good somatic growth. Only two patients had stunting, both of whom had recurrent infections prior to surgery and one had delayed surgery, because of parental ignorance.

Clinico-radiological assessment of musculoskeletal abnormality in relation to type of incision is as shown in table II.

Table 2:Distribution of musculoskeletal abnormalities in relation to thoracotomy incision:

Type of Incision	Muscle Cutting	Muscle Sparing
1. Chest wall deformity	1	1[a]
2. Ipsilateral Winging of Scapula [Figure 3]	5	2
3. Ipsilateral Rib deformity [b]	4	3
- Sub periosteal reaction	3	2
- Rib fusion	0	1
- Rib fracture	1	0
4. Ipsilateral Rib crowding	4	2
5. Scoliosis	0	1[c]
6. Ipsilateral Plural fibrosis	4	0

a- Case 14; antero-lateral chest wall depression associated with preoperative ICD insertion.[Figure 2]

b- Ipsilateral Rib deformity includes- Sub periosteal reaction, rib fusion and rib fracture.

c- Seen only on Chest X ray; Cobb's angle 13°.[Figure 4]

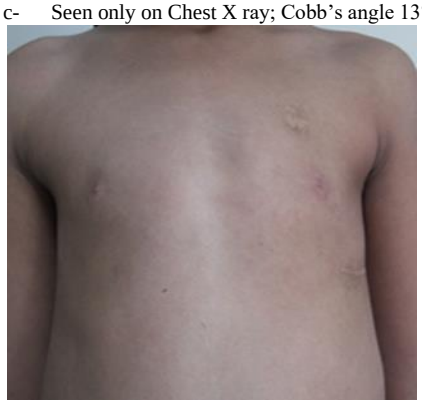

Fig 2: Showing left antero- lateral chest wall depression

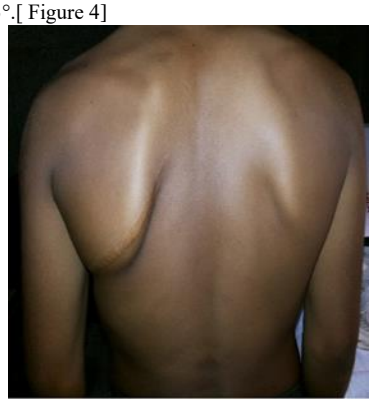

Fig 3:Showing winging of ipsilateral (left) scapula

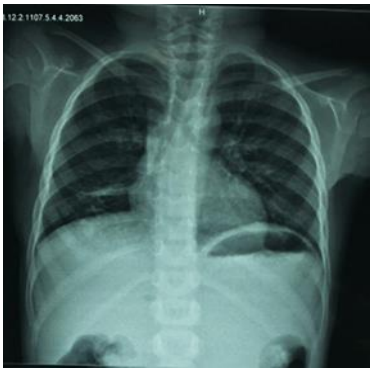

Fig 4:Showing scoliosis

All cases had shown compensatory growth of the remaining lung tissue, irrespective of the age at surgery and the indication for surgery.

Discussion

It has been observed that pulmonary resection in childhood, either congenital or acquired lesions, is well tolerated with a good somatic growth. [1, 9, 10, 11] Exceptionally poor body growth has been reportedin few but recurrent .[12]Kamata et al. reported a significant restriction in growth of the children with prenatally diagnosed cystic lung disease.[12] They concluded that fetal hydrops, lobectomy, CCAM, mediastinal shift and frequent respiratory infections significantly restrict the body growth. Pinter et al. [13] reported a better growth in patients operated for CCAM in later childhood compared with those operated in infancy. They postulated that CCAM has a worse natural history and prognosis if it manifests in early infancy. It seems likely that the poor outcome in infancy is related to the natural history of the disease itself rather than the procedure. We found no growth restriction in 90% of our patients. Only two patients (case 17 and 14) had stunted growth, at 1 and 1.5 year of follow up respectively. One of them (case 17) had been operated for post tubercular bronchiectasis, while the other (case 14) for CCAM. Both had recurrent pulmonary infections prior to surgery, which might be a reason for growth restriction in these cases. Malnutrition or constitutional stunting can also account for restricted growth. Also there remains a possibility that patients may catch up on their physical growth with passage of time. In our study, we have assessed their growth at only one point that too quiet early (1 and 1.5 years respectively). Therefore, it is unreasonable to attribute relative growth failure in these cases to the disease process and almost certainly to the procedure.

Exercise capacity in children following lung resection has been rarely evaluated. Eren et al. [10] and Frencker et al.[14] have reported normal exercise tolerance in children following lung resection for acquired as well as congenital lesions. While, Sritippayawan et al. [15] demonstrated exercise intolerance in about (20%) children undergoing lung resection, but they could not ascertain the factors responsible for such observations. None of our patients except one (case 8), operated for CCAM at an age of 2.5 month, complained of early fatigability on routine physical activities. This patient was assessed at 4.8 years of follow up. We admit that our assessment was subjective as we did not perform any objective test to evaluate the exercise capacity of the child. This child had multiple readmissions postoperatively for upper respiratory infections, and at present is under observation of the paediatrician for management of hyper-reactive airway. There have been recorded improvements when the child had been put on bronchodilators. It indicates that co-morbidities may be responsible for poor exercise tolerance. The original lung pathology and the operative procedure for the same cannot be held responsible for poor exercise tolerance.

That the procedure of thoracotomy itself is associated with postoperative musculoskeletal deformities is well known.[3, 16-18] The muscle cutting posterolateral thoracotomy incision may result in long-term physical impairment through musculoskeletal abnormalities. [3] All our cases were operated by postero lateral thoracotomy approach (60% muscle cutting and 40% muscle sparing). Ninety percent of our patients did not have any chest wall deformity and 65% did not show winging of scapula. None of the patient had scoliosis on clinical examination. Scoliosis is known to be associated with vertebral anomalies, but our selection process had excluded patients with known vertebral anomalies.

Several series have reported lower incidence of musculoskeletal abnormalities following muscle sparing thoracotomy**.[**19-21]In our study we also found similar association. There were lower musculoskeletal abnormality (winging of scapula, rib deformity, rib crowding and pleural fibrosis) in patients having muscle sparing incision.

Majority of the existing literature on outcome of lung resection in children has shown that the remaining lung grows well and compensate for the removed portion, irrespective of the indication. [10, 22-25]Findings to the contrary has also been reported in CLL.[26]However, age at surgery has been shown to have an effect on lung growth after lung resection.[27]It seems likely that in the earlier years of life it's the multiplication of lung tissue which compensates for the resected portion but after the lung growth slows down, over inflation of the alveoli takes over the role of compensation. Unlike adults this compensatory over inflation does not results in symptomatic emphysematous changes which can cause obstructive pulmonary dysfunction in children. [28]Therefore they had recommended the resection of lung cysts within the first two years of life. As no age related complication is seen it can be recommended that CLL can be resected early in life. The optimal age for surgical intervention is early infancy, to have good compensatory growth and better pulmonary function outcome.[27-29]Eren et al. had also reported marked herniation of the remaining lung in the patients of pneumonectomy, with a mediastinal shift to opposite side, on a median follow up of 5.2 years (ranged from 1 to 12 years).[10]

In our study, majority of the patients who underwent lung resection (lobectomy /pneumonectomy) had compensatory expansion of the remaining lung tissue on chest x ray, except one, who had shown only partial expansion of the remaining ipsilateral lobe. Herniation of the remaining lung seems to be most marked after pneumonectomy.

Previous studies, although scant, documented variety of deficits in pulmonary function, depending on the methodology used (spirometry, body plethysmography, ventilation/perfusion scan). Most reports have described normal function after surgery. Nakajima et al. in their study have concluded that the prognosis of PF after lobectomy for cystic lung disease in childhood was satisfactory.[28] Ayed et al. had also observed no alteration in pulmonary function in children who underwent lung resection for infectious pulmonary diseases, since the resected segment did not contribute to ventilation.[25] On the contrary, in a series of 13 patients, Sritippayawan et al. (2012),had demonstrated abnormal lung function after surgery in 76.9% (n=10) of their patients.[15]

As literature [30]had suggested that even preschool children (younger than 7 years of age) can perform spirometry, all our patients were subjected for spirometric assessment. However only 9 out of 20 patients were able to perform the test (Median age 11 years; 6-14 years). All these patients (100%) had an abnormal lung function test. We have used muscle cutting incision in all these patients, which is already an established factor for abnormal pulmonary function. [19] However, because the numbers are small, we can't assume an association with muscle cutting incision and abnormal PFT. Further the abnormal PFT may be due to some co-morbidity, and therefore reversible.

In six (66.7%) out of the nine patients. This is similar to the previous studies.[4, 10, 15]Five of these patients (83.3%) had mild restrictive defect and one patient (16.7%) had moderate restriction, results being similar to others.[4]

Out of the 5 patients with mild restriction, two patients had had pneumonectomy for destroyed lung (case 10 and 20). Rib crowding and pleural fibrosis was present in these patients both of which may cause restrictive lung defects. The rest of the three patients had lobectomy. Two of them (case 6 and 19) were operated for acquired lung disease and one for CCAM (case 12).

The patient with CCAM(case 12) underwent right lower lobectomy at age of 12.25 years and had multiple infections prior to surgery. The age at operation has been shown to affect the prognosis of PF.[28]Younger the age at surgery better was the outcome. Komori et al. [27] had found better pulmonary function in children operated for CLL in infancy compared to those operated after one year of age while Nakajima et al. have defined the younger age group to be less than 4 years. Delayed referral may be the cause of poor pulmonary

function in the patient. Some studies did not find any correlation between age[31, 32]and postoperative lung function.

The patients with acquired lung disease had infected hydatid cyst (case 6) and recurrent LCH [Langerhans cell histiocytosis] (case 10), and underwent surgery at 7.5 and 9.6 years of age respectively. Recurrent infections may explain the restrictive pattern in these two patients. Also the infected group had older children compared to non-infected, a factor which can explain the poorer PF[Pulmonary function] outcome after surgery.The patient (case 7) with moderate restriction, had been operated twice for necrotising pneumonia. It has been established that recurrent pulmonary infections,[27, 28]multiple thoracotomies,[4]and muscle cutting incision has been associated with poor pulmonary function these can be the causes of moderate restriction in our patient. Isolated obstructive dysfunction was present in 22.22% of our cases similar to Sritippayawan[15] (20%), but both of our cases had reversible obstruction. It can be due to hyper reactive airway, and not consequent to lung resection. Panda et al. had not found any obstructive abnormality as respiratory symptoms of the patients were taken care of.[4]This again suggests that obstructive PFT need not be due to disease itself or the procedure. It could be due to a co-morbidity. Only one patient (case 9) had a mixed defect, severe restriction with partially reversible obstruction. Chronic airway disease is suspected to be a possible cause of this abnormality by Paediatrician and child was put on inhalational steroids. Sritippayawan also reported mixed restrictive and obstructive defect in one of their patient who had lobectomy for CCAM.We conclude that lung resection surgery is well tolerated by children , child attain good somatic growth and the remaining lung tissue grow to compensate , irrespective of the age and the indication for surgery, Musculoskeletal abnormalities are more following muscle cutting incision. Muscle sparing incision carried comparatively a good outcome. All these patients had shown an abnormality on PFT. Restrictive defect was the most common abnormality seen in our cases. Obstructive pattern was seen in few cases, but it was reversible, indicating that it was unrelated to surgery. The data on PFT suggests that assessment of PFT in post op period is useful to address co-morbidities that can cause functional impairment. The original condition and the surgical procedure may not be responsible for abnormal PFT. Hence, their longitudinal follow up is importance.

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Conflict of Interest: Nil
Source of support:Nil