

## Pulmonary sublobar resections in pediatric patients

### *Pediatric hastalarda pulmoner sublobar rezeksiyonlar*

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**Background:** This study aims to evaluate the outcomes of pulmonary sublobar resection in pediatric patients.

**Methods:** We retrospectively reviewed the medical records of 405 children who were operated in the Thoracic Surgery Clinic of İstanbul University Medical Faculty between January 2001 and June 2011. Only 43 patients (20 girls, 23 boys; mean age 11.4±0.5 years; range 0 to 16 years) who underwent pulmonary sublobar resection were included in the study. Patients were evaluated in terms of age, gender, complaints on admission, history of previous diseases, diagnostic methods, operative indications, surgical procedures, complications, duration of hospitalization and outcome of the procedure.

**Results:** Indications for pediatric resections were oncological (n=29), non-oncological (n=9) and congenital pathologies (n=5). The mean duration of hospitalization was 3.6 days after surgical procedure. Morbidity was observed in two patients postoperatively, whereas mortality was not observed.

**Conclusion:** Indications of pulmonary sublobar resections varies in pediatric patients compared to adults. Pulmonary sublobar resection is a good option for children, as less functioning lung tissue is removed with fewer postoperative complications. It is also relatively simple and fast surgical method, than lobectomy.

**Key words:** Pediatric; pulmonary sublobar resection; segmentectomy; wedge resection.

**Amaç:** Bu çalışmada, pediatrik hastalarda pulmoner sublobar rezeksiyonunun sonuçları değerlendirildi.

**Çalışma planı:** Ocak 2001 - Haziran 2011 tarihleri arasında İstanbul Üniversitesi İstanbul Tıp Fakültesi Göğüs Cerrahisi Kliniğinde ameliyat edilen 405 çocuğun tıbbi kayıtları geriye dönük olarak incelendi. Yalnızca pulmoner sublobar rezeksiyon uygulanan 43 hasta (20 kız, 23 erkek; ort. yaş 11.4±0.5 yıl; dağılım 0-16 yıl) çalışmaya dahil edildi. Hastalar yaş, cinsiyet, başvuru yakınmaları, geçirilmiş hastalık öyküsü, tanı yöntemleri, ameliyat endikasyonları, cerrahi yöntemler, komplikasyonlar, hastanede kalış süreleri ve işlemin sonuçları yönünden değerlendirildi.

**Bulgular:** Cerrahi rezeksiyon endikasyonları onkolojik (n=29), non-onkolojik (n=9) ve doğuştan patolojiler (n=5) idi. Cerrahi işlem sonrası ortalama hastanede yatış süresi 3.6 gün idi. Ameliyat sonrası iki hastada morbidite görülür iken, mortalite görülmedi.

**Sonuç:** Pulmoner sublobar rezeksiyonların çocuk hastalardaki endikasyonları yetişkinlere göre farklılık gösterir. Fonksiyonel akciğer dokusu kaybının ve ameliyat sonrası komplikasyonların daha az olması nedeni ile, pulmoner sublobar rezeksiyon çocuklar için iyi bir seçenektir. Ayrıca, lobektomiden daha kolay ve hızlı yapılabilir cerrahi bir yöntemdir.

**Anahtar sözcükler:** Çocuk; pulmoner sublobar rezeksiyon; segmentektomi; kama rezeksiyonu.



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Since the anatomical and physiological features of pediatric patients are different, they are more sensitive to postoperative problems.<sup>[1,2]</sup> Therefore, they should be closely followed up in the postsurgical period. The patient's blood pressure, heart rate, pulmonary functions, peripheral oxygen saturation, blood gases, electrocardiogram, and temperature should be monitored carefully, and invasive measures should be taken when necessary.<sup>[3]</sup> In the literature, there is limited data regarding sublobar resections in pediatric patients. Hence, the purpose of this study was to evaluate pediatric patients who underwent pulmonary sublobar resections for oncological, non-oncological, and congenital indications.

## PATIENTS AND METHODS

Between January 2001 and June 2011, a total of 405 pediatric patients were subjected to surgical intervention at the İstanbul University Medical Faculty, Department of Thoracic Surgery. Those who underwent a lobectomy or a pneumonectomy were excluded from the study while the 43 for whom a pulmonary sublobar resection was performed were included. The medical records of these patients (23 males, 20 females; median age 11.4±0.5 years; range 0 to 16 years) were then retrospectively evaluated in terms of age, gender, complaints, history of disease, diagnostic methods, operative indications, surgical procedures, complications, duration of hospitalization, and outcome.

## RESULTS

Twenty-two patients (51.2%) had no symptoms related to pulmonary pathology. Coughing (with or without expectoration or fever) (25.5%) was the most common complaint, followed by chest pain (14.0%), shortness of breath (7.0%), and fever alone (2.3%). In addition, the majority of the patients had no history of disease; however, a history of cancer was noted in 26 patients (60.5%) (Table 1). As a diagnostic determinant, a plain chest X-ray was performed on all of the patients. Other diagnostic methods were also used in this study including computed tomography (CT) for 30 patients (69.8%), CT + positron emission tomography (PET) for five others (11.6%), high resolution computed tomography (HRCT) for five more (11.6%), CT + magnetic resonance imaging (MRI) for two (4.6%), CT + Doppler ultrasonography (US) for one (2.3%).

The indications of pediatric pulmonary resections were grouped into three categories: oncological, non-oncological, or congenital pathologies. The

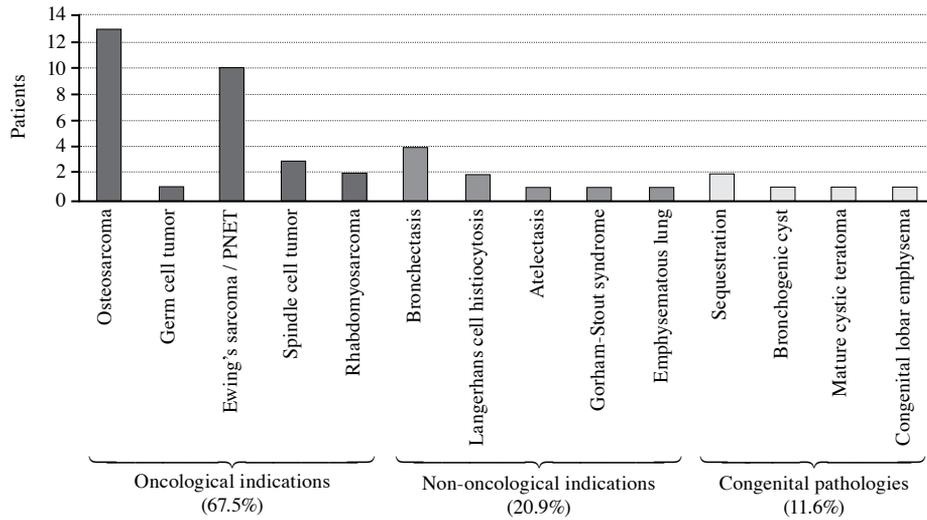
most frequent indication was oncological pathologies (n=29, 67.5%), with pulmonary metastasis of osteosarcoma (30.2%) being seen the most. Non-oncological and congenital indications were observed in nine (20.9%) and five (11.6%) patients, respectively, with bronchiectasis being the most frequent non-oncological pathology (9.3%) and sequestration occurring most often in the congenital category (4.6%) (Figure 1).

In terms of surgery, 45 segmentectomies + wedge resections (7 right upper, 9 right lower, 16 left upper, 9 left lower, and 2 bilateral) were performed on 43 patients. Out of these, five were performed via video-assisted thoracic surgery (VATS). In addition, a bilateral upper + lower lobe segmentectomy + wedge resection was performed on one patient. Furthermore, a right thoracotomy was performed on 16 patients, a left thoracotomy on 21, a bilateral thoracotomy on one, a right thoracotomy + left VATS on one, and VATS alone on four others (Figure 2).

First, the segmentary artery and bronchi were isolated, ligated, and divided during the segmentectomy. Then the distal part of the segmental bronchi was occluded with a clamp, and the atelectatic segment was removed from the expanded lung with blunt dissection along the intersegmentary vein. This vein was kept intact with its main branches because they supply the two segments which were left behind.

**Table 1. The clinical features of the patients (n=43)**

	n	%
Gender		
Male	23	53.5
Female	20	46.5
Complaints		
None	22	51.2
Fever	1	2.3
Cough	4	9.3
Cough + fever	3	7.0
Cough + expectoration	4	9.3
Shortness of breath	3	7.0
Chest pain	6	14.0
History of disease		
None	14	32.6
Operated osteosarcoma	13	30.2
Operated Ewing's sarcoma	7	16.3
Spindle cell tumor	3	7.0
Operated rhabdomyosarcoma	2	4.7
Frequent lung infection	1	2.3
Bronchitis	1	2.3
Germ cell tumor	1	2.3
Tonsillectomy	1	2.3



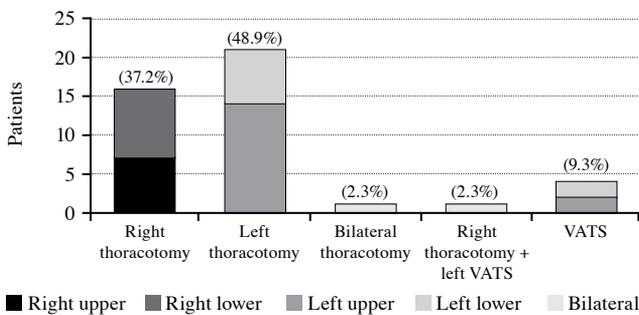
**Figure 1.** Indications for pediatric pulmonary sublobar resections. PNET: Primitive neuroectodermal tumor.

The average duration of hospitalization was 3.6 days. Postoperative morbidity was observed in two patients who underwent rib resections in our study, whereas there was no mortality. The re-expansion defect was also observed in one patient (2.3%), and fever was seen in six (13.8%) (Figure 3).

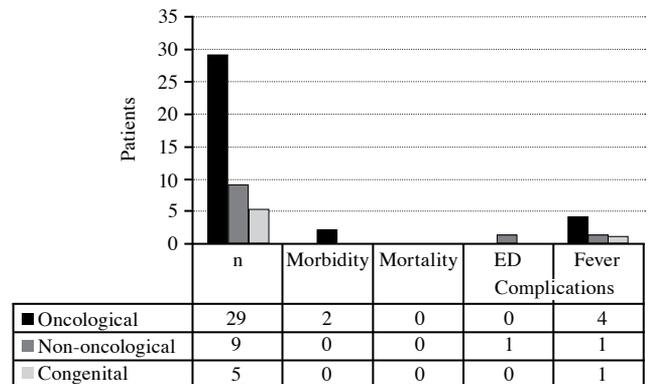
**DISCUSSION**

A pulmonary sublobar resection (a segmentectomy or wedge resection) is defined as the removal of a segment or a part of a lung lobe. This method is beneficial in that it preserves lung function well, and it is mostly reserved for adult patients with a poor cardiopulmonary reserve. In addition, sublobar resections are important because they preserve pulmonary function in pediatric patients with pulmonary pathologies. To the best of our knowledge, although this procedure has been performed for decades, there is limited data regarding pediatric sublobar resections in the literature, and our

study appears to be one of the few that have focused on this topic. The indications for thoracic surgery vary in children. In this study, the pediatric patients who underwent a pulmonary sublobar resection were placed in categories according to whether they had oncological, non-oncological, or congenital pathologies, with the most frequent being oncological. In addition, all of the patients had metastatic lesions in their lungs. Cohen and Kaschula<sup>[4]</sup> reported that the ratio of pediatric primary pulmonary neoplasms to benign lesions or developmental masses was 1:10 while the percentages of primary tumors, metastatic tumors, and non-neoplastic lesions were 1, 5, and 60%, respectively. A complete resection with a lobectomy or pneumonectomy is the standard treatment for primary lung tumors, and a sublobar resection is associated with increased local recurrence. Thus, all of the patients in our study with a primary origin underwent either a lobectomy or pneumonectomy but



**Figure 2.** Surgical procedures for pediatric pulmonary sublobar resections. VATS: Video-assisted thoracic surgery.



**Figure 3.** Morbidity, mortality, and complication rates. ED: Expansion defect.

not a sublobar resection, and those with primary lung tumors were not included. Furthermore, we found that the percentage of primary tumors to metastatic tumors occurred at a ratio of 1:5, which was in accordance with the literature (data not shown). Metastatic tumors constituted the majority of the cases in our study. Perhaps this was because the patients with non-oncological and congenital pathologies were operated more often at secondary level hospitals while the patients with oncological pathologies were referred to our facility since it is a tertiary level hospital.

It can be difficult to diagnose metastatic lung tumors in pediatric patients because they may mimic more common conditions and sometimes present with non-specific symptoms such as coughing, pneumonia, hemoptysis, and shortness of breath. However, most of the tumors can be detected during follow-up examinations.<sup>[5]</sup> Although coughing was the most common symptom in our study, the vast majority of the patients had no symptoms related to pulmonary pathology, resulting in delayed diagnoses and treatment. Our findings suggest that metastatic lung tumors may be asymptomatic; therefore, physicians should remain alert for metastasis in the lungs during the follow-up of patients with extrapulmonary primary malignancies. The most common metastatic lung tumors in the children in our study were osteosarcoma and Ewing's sarcoma. Some patients had more than one metastatic lesion, and most had metastatic tumors located in the peripheral lung. Currently, there is no standardized treatment for resecting pulmonary metastases among children, and differences can be seen with regard to the extent of the resection (segment, wide wedge, or close wedge) and the approaches used in VATS and thoracotomies. A peripherally located solitary lesion of less than 3 cm in diameter is the main indication for a sublobar resection. However, a lobectomy is preferred when the lesion is centrally located, regardless of its diameter.

Recent studies have shown that aggressive surgical approaches to pulmonary metastases in osteosarcoma improve the rate of survival.<sup>[6,7]</sup> Meyers et al.<sup>[8]</sup> found a correlation between complete resection of the primary tumor and all metastases with general survival rates in osteocarcinoma patients with synchronized pulmonary metastases. In a study by Letourneau et al.<sup>[9]</sup> that involved patients with lung metastases due to Ewing's sarcoma, they compared those who underwent a metastasectomy with those who did not and reported that those who had surgery had a better overall survival rate. Our findings were similar in that they showed that metastasectomies improved the chance of survival.

Thus, we suggest that a metastasectomy can be a lung-saving procedure for pediatric patients with metastatic disease.

Karnak et al.<sup>[10]</sup> reported that a pulmonary metastasectomy might increase the rate of survival in carefully selected children, but that it was unlikely to cure them. Therefore chemotherapy and/or radiotherapy needs to be continued in the postoperative period, and the appropriate patients received these therapies in our study.

In addition, we believe that surgery combined with oncological treatment for metastatic disease prevents recurrence and improves the chance of survival. In our study, 20.7% of the patients underwent surgery due to non-oncological indications, and bronchiectasis was seen the most often. The first treatment for this disease state is a conservative one in which antibiotics, postural physiotherapy, bronchodilators, and corticosteroids are prescribed to reduce airway obstruction and eliminate lower airway microorganisms.<sup>[11]</sup> However, we preferred and suggested that a segmentectomy be performed procedure for those patients with localized bronchiectasis. Other indications for non-oncological pathologies in our study were Langerhans cell histiocytosis, atelectasis, Gorham-Stout syndrome, and an emphysematous lung. A wedge resection was performed to accurately diagnose the patients with Langerhans cell histiocytosis. Gorham-Stout syndrome (vanishing bone disease) is a rare disorder characterized by spontaneous bone resorption. It progresses rapidly and occasionally occurs after minor trauma. This is usually followed by thoracic cage, pulmonary, or pleural involvement, which leads to a compromise of respiratory function and subsequent death.<sup>[12]</sup> We performed a wedge resection in a patient with Gorham-Stout syndrome who had damaged lungs as the result of a chylothorax. We also performed a wedge resection or segmentectomy on appropriate patients with atelectasis or emphysematous lung disease.

In our study, 11.6% of the patients underwent a sublobar resection due to congenital pathologies such as sequestration, a bronchogenic cyst, a mature cystic teratoma, and congenital lobar emphysema, and no recurrence was seen in any of these patients. Hence, a sublobar resection is advocated for these patients when possible.

Pediatric patients usually undergo direct X-rays first step in the investigative process. Although direct posteroanterior (PA) X-rays can be useful for lung and heart problems, vascular interstitial pathologies, especially in the thoracic wall, may require advanced

investigations such as CT, MRI, or angiography.<sup>[13]</sup> In our study, all of the patients had direct chest X-rays while 30 (69.8%) also underwent CT, five (11.6%), had HRCT, five (11.6%) had CT + PET, two (4.6%) underwent CT + MRI and one (2.3%) was prescribed CT + Doppler US. We believe that preoperative CT is very useful for evaluating metastatic lung lesions and for determining which lung segments should be resected. Because some small lesions may be missed on CT, surgeons should always palpate the lungs via gentle superficial palpation to eliminate the possibility of missed lesions during surgery.

A muscle-sparing lateral thoracotomy provides sufficient exposure for pediatric patients, and VATS is a minimally invasive method that has recently been used in conjunction with thoracic surgery. In our study, left upper and lower segmentectomies + wedge resections were performed on four patients via VATS. We routinely perform lobectomies and/or sublobar resections using VATS on adults and desire to increase our use of VATS with children. In a study by Findik et al.<sup>[14]</sup> in which pediatric patients were reviewed for thoracotomies, complications were observed in 35 patients (18%), with atelectasia and secretion retention in 54%, postoperative extended air leakage in 20%, wound infections in 17%, hemorrhage in 3%, chylothoraces in 3%, and intrathoracic space in another 3%. In our study, morbidity was seen in two cases who underwent a costal resection together with a pulmonary sublobar resection due to Ewing's sarcoma, but no mortality occurred. Additionally, postoperative complications were observed in seven of our patients (16.1%), with fever in six (13.9%) and an expansion defect in one (2.3%). All of the complications were remedied by close follow-up and medical treatment.

### Conclusion

Pediatric pulmonary sublobar resections have different indications with respect to adults, but we believe that they are a good option for children because less functioning lung tissue is removed in this procedure, and there are fewer postoperative complications. Furthermore, this type of resection is simpler and faster than a lobectomy. Therefore, in pediatric patients, a pulmonary sublobar resection should be preferable to either a lobectomy or pneumonectomy since it is a lung-saving procedure that can be used in the management of peripheral metastatic lesions or other non-oncological conditions of the lungs. In addition, this procedure provides a successful outcome with low morbidity and mortality along with fewer complications when performed at experienced medical centers.

### Declaration of conflicting interests

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