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Pulmonary sublobar resections in children with congenital cystic adenomatoid malformations

Doğuştan kistik adenomatoid malformasyonlu çocuklarda pulmoner sublober rezeksiyonlar

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ABSTRACT

Background: In this study, we aimed to evaluate the outcomes of pulmonary sublobar resections performed for congenital cystic adenomatoid malformations (CCAM) in pediatric patients.

Methods: We retrospectively reviewed 20 children (11 boys, 9 girls; mean age 5.48 years; range 39 days to 18 years) who underwent surgery for CCAM in our clinic between January 1999 and December 2012. Eleven patients had lobectomies (group 1), and nine patients had sublobar resections of whom six underwent thoracoscopic resections and three underwent open segmentectomy (group 2). Data were collected regarding age, sex, pathological diagnosis, location of lesion, surgical procedure, size of the lesion, chest tube duration, length of hospital stay, and postoperative complications.

Results: Although there was no statistically significant difference in the demographics between the two groups undergoing lobar and sublobar resection, the chest tube duration (5.3±0.5 days vs 3.6±1.9 days) and the length of hospital stay (7.5±0.7 vs 4.8±2.2 days) were significantly shorter in the sublobar resection group ($p<0.05$).

Conclusion: In our experience, both lobar and sublobar resections have similar clinical outcomes. Sublobar resections can be, therefore, easily performed thoracoscopically and can be safely applied to pediatric patients with CCAM.

Keywords: Congenital cystic adenomatoid malformation, sublobar resection; thoracoscopy.

ÖZ

Amaç: Bu çalışmada pediatrik hastalarda doğuştan kistik adenomatoid malformasyonlar (DKAM) nedeni ile yapılan pulmoner sublober rezeksiyonların sonuçları değerlendirildi.

Çalışma planı: Ocak 1999 - Aralık 2012 tarihleri arasında kliniğimizde DKAM nedeni ile cerrahi uygulanan 20 çocuk (11 erkek, 9 kız; ort. yaş 5.48 yıl; dağılım 39 gün - 18 yıl) retrospektif olarak incelendi. On bir hastaya lobektomi (grup 1) ve dokuz hastaya altısı torakoskopik rezeksiyon ve üçü açık segmentektomi olmak üzere sublober rezeksiyon (grup 2) yapıldı. Yaş, cinsiyet, patolojik tanı, lezyonun yeri, cerrahi işlem, lezyonun büyüklüğü, göğüs tüpü süresi, hastanede kalış süresi ve cerrahi sonrası komplikasyonlara ilişkin veri toplandı.

Bulgular: Lobar ve sublober rezeksiyon yapılan iki grup arasında demografik açıdan istatistiksel olarak anlamlı fark olmamasına rağmen sublobar rezeksiyon yapılan grupta göğüs tüp kalış (sırası ile 5.3±0.5 gün, 3.6±1.9 gün), hastanede kalış süresi (sırası ile 7.5±0.7, 4.8±2.2) anlamlı olarak azalmıştı ($p<0.05$).

Sonuç: Deneyimimize göre, hem lober hem de sublober rezeksiyonların klinik sonuçları benzer idi. Bu neden ile sublober rezeksiyonlar torakoskopik olarak kolaylıkla gerçekleştirilebilir ve DKAM'lı pediatrik hastalara güvenli bir şekilde uygulanabilir.

Anahtar sözcükler: Doğuştan kistik adenomatoid malformasyon, sublober rezeksiyon; torakoskopi.

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Congenital lung malformations (CLMs) include several abnormalities, such as congenital cystic adenomatoid malformations (CCAMs), pulmonary sequestrations, congenital lobar emphysema, and peripheral bronchogenic cysts.[1] Congenital cystic adenomatoid malformations are rare anomalies of the lower respiratory system. Until recently, lobectomy was the cornerstone of therapy for these congenital lung lesions.[2] However, minimally invasive surgery for these malformations has been widely adopted by pediatric surgeons over the past decade. On the other hand, it is associated with increased morbidity risks caused by the thoracotomy incision in a growing child. These risks include shoulder girdle weakness, chest deformities, and scoliosis.[3]

Over the past decade, the video-assisted thoracoscopic surgery (VATS) which is an alternative to open procedures has yielded excellent results.[4] Meanwhile, sublobar parenchymal-saving resections have been also increasingly adopted.[5] Sublobar resections include resections containing part of a lobe (segmentectomy or wedge resection). Sublobar resections can be successfully performed in CCAMs via thoracoscopy.[6] Review of the literature revealed limited data on the sublobar resections of CCAMs in pediatric patients. Herein, we examined the efficacy of sublobar resections performed for CCAMs and discussed a series of lung resections for CCAMs performed in a single center.

PATIENTS AND METHODS

We retrospectively reviewed the records of 20 children (11 boys, 9 girls; mean age 5.48 years; range 39 days to 18 years) admitted to Şişli Etfal Education and Research Hospital, Department of Pediatric Surgery, who underwent pulmonary resections for CCAMs between January 1999 and December 2012. Age at operation, sex, localization, type of procedure, pathological diagnosis, length of hospital stay, time of chest tube placement, and postoperative complications were retrospectively reviewed. Eligibility for inclusion was determined by the pathological result of the surgical specimen. All other congenital lung lesions except CCAM were excluded. All cases underwent either lobectomy or segmentectomy for CCAM.

Among 20 patients, group 1 consisted of 11 patients who underwent lobectomy, while group 2 consisted of nine patients who underwent sublobar resections (six thoracoscopic resections, three open segmentectomy resections).

All patients underwent preoperative computed tomography (CT). The patients with open resection underwent standard muscle-sparing posterolateral thoracotomy, as outlined elsewhere.[3] First, the segmental artery and bronchi were ligated and the segment with the lesion was, then, removed. Thoracoscopic operations were performed with selective lung intubation via three ports. Tissue divisions were adequately carried out with the combined use of a sealing device (LigaSure LS1000; Covidien) and rotaculator endoscopic staplers (Endo GIA Ultra Universal Stapler, Covidien Surgical, Mansfield, MA, USA). Outcome variables included the length of hospital stay, chest tube duration, and postoperative complications.

A routine postoperative CT was not performed on any of the patients, unless there was a respiratory problem and a request during the annual follow-up visit with plain chest radiography. The mean follow-up was 94.8 months (range 30 to 186 months).

Statistical analysis

The statistical analysis of the data was performed by using the SPSS for Windows version 16.0 software program (SPSS Inc., Chicago, IL, USA). The Fisher's exact test and Mann-Whitney U test were performed to analyze abnormally distributed data. A 95% confidence interval was accepted to be the confidence interval and a p value of ≤ 0.05 was considered statistically significant.

RESULTS

Data related age, sex, location and size of the lesion, surgical procedure, and pathological diagnosis are shown in Table 1. Age at the time of surgery ranged from 39 days to 18 years. The mean and median age were 5.48 years and one year, respectively. According to the pathological reports, 11 patients had type 1 CCAM lesions, whereas seven patients had type 2 CCAM lesions. There were no significant differences in the mean age, sex, or localization of resection between the groups ($p>0.05$) (Table 2).

Primary postoperative outcome variables, namely chest tube duration and length of hospital stay are also listed in Table 1. We found an increased chest tube duration and prolonged hospital stay in the lobectomy group, indicating a significant difference ($p=0.045$, $p=0.021$, respectively). No mortality or short-term complications were observed.

Recurrence was not observed in any of the patients during follow-up. Computed tomography results were able to be evaluated in only three patients who were admitted with lower respiratory system infection findings in the late period; these infections

Table 1. Age, sex, location and size of lesion, access, surgical methods, and pathological diagnosis

Age (year)	Sex	Location	Access	Surgery	Size	Pathological diagnosis
0.83	F	RLL	Thoracotomy	Lobectomy	5x5x7	Type 1
3.5	M	LLL	Thoracotomy	Lobectomy	8x6x4	Type 1
1	M	LLL	Thoracotomy	Lobectomy	5x6x7	Type 1
0.083	M	RUL	Thoracotomy	Lobectomy	5x5x6	Type 2
0.083	F	RML	Thoracotomy	Segmentectomy	3x4x5	Type 1
11	M	RLL	Thoracotomy	Lobectomy	8x7x3	Type 1
0.083	F	LLL	Thoracotomy	Segmentectomy	5x4x3	Type 1
0.41	F	LUL	Thoracotomy	Lobectomy	6.5x5x1.5	Type 2
10	M	RUL	Thoracotomy	Lobectomy	5.5x4x2	Type 1
0.75	M	RLL	Thoracotomy	Lobectomy	1x0.6x3	Type 1
1	F	RML	Thoracotomy	Segmentectomy	5x3x1	Type 1
8	F	LUL	Thoracotomy	Lobectomy	15x10x2.5	Type 1
11	F	LLL	Thoracotomy	Segmentectomy	13x11x5.5	Type 2
0.5	F	RLL	Thoracoscopy	Segmentectomy	5x4x3	Type 1
0.33	M	RML	Thoracotomy	Segmentectomy	6.5x4.5x2.5	Type 1
0.083	M	LUL	Thoracotomy	Lobectomy	10x8.5x1.5	Type 2
18	F	RLL	Thoracotomy	Lobectomy	11.5x7x5	Type 2
12	M	RLL	Thoracoscopy	Segmentectomy	5.5x2.5x1.7	Type 1
14	M	RML	Thoracoscopy	Lobectomy	5x5x6	Type 2
17	M	RUL	Thoracoscopy	Segmentectomy	10x15x20	Type 2

RLL: Right lower lobe; LLL: Left lower lobe; RUL: Right upper lobe; RML: Right middle lobe; LUL: Left upper lobe.

resolved with antibiotherapy. Two of these patients were lobectomy patients, while the other one had segmentectomy. No patients required second-look surgery.

DISCUSSION

Thanks to the advancements in imaging techniques, the detection of CCAM has increased.[7,8] In recent years, elective and lung-sparing surgical techniques have been preferred, although the lesions were previously treated with lobectomy in the presence of complications. Our clinic's approach has also changed with this trend. If applicable, we preferred VATS segmentectomies in our cases. In this study, we retrospectively reviewed our CCAM cases over 13 years and compared the sublobar resections with

Table 2. Characteristics and perioperative outcomes of patients who had lobar versus sublobar resections

	Variable lobar (n=11)				Sublobar (n=9)				
	n	Mean±SD	Median	Min.-Max.	n	Mean±SD	Median	Min.-Max.	*p*
Age (years)		6.0±4.9	1	0.083-18		6.2±7.1	1	0.083-17	0.88
Gender									
Male	7				4				
Female	5				5				
Anatomic localizations									
LLL	2				2				
LUL	3				–				
RLL	4				1				
RML	1				4				
RUL	2				1				
Chest tube duration (day)		5.3±0.5	5	5-6		3.6±1.9	3	2-7	0.045*
Length of hospital stay (day)		7.5±0.7	7	7-9		4.8±2.2	4	3-8	0.021*

SD: Standard deviation; Min.: Minimum; Max.: Maximum; LLL: Left lower lobe; LUL: Left upper lobe; RLL: Right lower lobe; RML: Right middle lobe; RUL: Right upper lobe; * Statistically significant.

lobectomies. In addition, one of the advantages of lung-sparing surgery is that it can be performed easily with thoracoscopy. Of note, prenatal diagnosis and early surgery are recommended to obtain the most successful results.[6]

Many authors used lung-sparing surgery to preserve lung tissue and to reduce morbidity eventually and reported that the early and late outcomes of the parenchyma-saving operations were excellent. Unlike our study, many of these surgeries included not only CCAMs, but also other CLMs, bronchopulmonary sequestration, bronchogenic and foregut cysts, bronchial atresia with distal cystic degeneration, and congenital lobar emphysema.[9-13]

The most common type of CCAM is type 1. In our series, the cases were either type 1 or 2. Type 1 cysts are larger than 2 cm and can be as large as 10 cm. These cysts are associated with malignant transformation.[14] One of the main reasons for elective resection for these lesions is their potential malignant nature. Therefore, all lesions are treated surgically, whereas they were treated conservatively and operated in the presence of complications in the past. We believe that the remaining tissue has a potential of development of malignancy.

Many studies have shown no differences between thoracotomy and thoracoscopy in terms of surgical outcome; however, thoracoscopy carries the advantages of being minimal invasive surgery, cosmesis, and muscle preservation in a growing child.[15-18] In the present study, three patients underwent thoracoscopic segmentectomies and we believe that segmentectomy in combination with thoracoscopy is a promising treatment of choice for the surgical management of CCAMs. Thanks to its lung-sparing potential, it may be convenient for thoracoscopy.

The main limitation of our study was the relatively small sample size. However, the types of lesions we studied are rare and all of our patients were pathologically diagnosed with CCAMs. Another limitation of our study was that our patients were unable to be analyzed for postoperative analgesia. Otherwise, we might possibly have proven that VATS yielded much better outcomes in postoperative analgesia.

In conclusion, parenchyma-sparing sublobar resections are appropriate alternatives which can be performed thoracoscopically and shorten the chest tube duration and length of hospital stay. We believe that lung-sparing surgery as sublobar resection may be sufficient in the treatment of CCAMs.

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