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Original Article / Özgün Makale

The role of current surgical therapy for pediatric/adolescent and adult patients with bronchiectasis

Bronşektazili çocuk/ergen ve erişkin hastalarda mevcut cerrahi tedavinin yeri

Hakkı Ulutaş, Muhammet Reha Çelik, Akın Kuzucu

Department of Thoracic Surgery, University of Inonu Faculty of Medicine Turgut Ozal Medical Center, Malatya, Turkey

ABSTRACT

Background: This study aims to compare clinical manifestations, surgical indications, surgical procedures, postoperative complications, and outcomes of surgical treatment in pediatric and adult patients with bronchiectasis.

Methods: Between January 2000 and December 2013, a total of 99 patients (40 males, 59 females; mean age 33.3 years; range 10 to 67 years) who underwent surgery for bronchiectasis at the Department of Thoracic Surgery were retrospectively analyzed. Group 1 (n=28) were pediatric patients (≤18 years) and group 2 (n=71) were adult patients (>18 years). Clinical symptoms, criteria surgical treatments applied, surgical treatment approaches, postoperative complications, the length of hospital stay, and treatment outcomes were evaluated.

Results: In both groups, the most common symptom was productive cough, followed by recurrent infections. There were no significant differences in the frequency of these symptoms between the groups. In total, 104 surgeries were performed. Of 29 surgeries in group 1, 26 were thoracotomies and three were video-assisted thoracoscopic surgery. Of 75 surgeries in group 2, 60 were thoracotomies and 15 were video-assisted thoracoscopic surgery. Postoperative complications were seen in eight operations (27.6%) in group 1 and 18 operations (24%) in group 2. There was no intra- or postoperative mortality. All 28 patients in group 1 showed improvement after surgery, while 23 outcomes (82.1%) were rated excellent (complete remission) and five outcomes (17.9%) were rated improved (reduced symptoms). In group 2, 63 outcomes (88.7%) were excellent and seven outcomes (9.9%) were rated improved; however, one patient (1.4%) showed no improvement.

Conclusion: In selected cases, surgical treatment for bronchiectasis is satisfactory and associated with acceptable mortality and morbidity rates, irrespective of the age of the patient. Based on our study results, surgery can cure this condition and can improve the quality of life, even when not curative.

Keywords: Adult bronchiectasis; pediatric bronchiectasis; surgical treatment.

ÖZ

Amaç: Bu çalışmada, bronşektazili çocuk ve erişkin hastalarda klinik bulgular, cerrahi endikasyonlar, cerrahi işlemler, ameliyat sonrası komplikasyonlar ve cerrahi tedavi sonuçları karşılaştırıldı.

Çalışma planı: Ocak 2000 - Aralık 2013 tarihleri arasında bronşektazi nedeni ile Göğüs Cerrahisi bölümünde ameliyat edilen toplam 99 hasta (40 erkek, 59 kadın; ort. yaş 33.3 yıl; dağılım 10-67 yıl) geriye dönük olarak incelendi. Grup 1 (n=28) çocuk hastalardan (≤18 yıl) ve grup 2 (n=71) erişkin hastalardan (>18 yıl) oluşuyor idi. Klinik semptomlar, yapılan cerrahi tedavi kriterleri, cerrahi tedavi yaklaşımları, ameliyat sonrası komplikasyonlar, hastanede kalış süreleri ve tedavi sonuçları değerlendirildi.

Bulgular: Her iki grupta en yaygın semptom prodüktif öksürüğü takiben tekrarlayan enfeksiyonlar idi. Gruplar arasında bu semptomların görülme sıklığı açısından anlamlı bir fark saptanmadı. Toplamda 104 cerrahi yapıldı. Grup 1'de 29 cerrahinin 26'sı torakotomi ve üçü video yardımlı torakoskopik cerrahi idi. Grup 2'de 75 cerrahinin 60'ı torakotomi ve 15'i video yardımlı torakoskopik cerrahi idi. Grup 1'de sekiz ameliyatta (27.6%) ve grup 2'de 18 ameliyatta (24%) ameliyat sonrası komplikasyonlar izlendi. Ameliyat sırası ve sonrası mortalite gözlenmedi. Grup 1'de 28 hastanın tamamında ameliyat sonrası iyileşme izlenir iken, 23 sonuç (%82.1) mükemmel (tam remisyon) ve beş sonuç (%17.9) iyileşmiş (semptomlarda azalma) olarak tanımlandı. Grup 2'de 63 sonuç (%88.7) mükemmel ve yedi sonuç (%9.9) iyileşmiş olarak tanımlandı; ancak bir hastada (1.4%) iyileşme izlenmedi.

Sonuç: Seçilmiş olgularda, hastanın yaşından bağımsız olarak, bronşektazinin cerrahi tedavisi tatmin edicidir ve kabul edilebilir mortalite ve morbidite oranları ile ilişkilidir. Çalışma sonuçlarımıza göre, cerrahi bu hastalığı tedavi edebilir ve tedavi edemediği durumlarda dahi, hastanın yaşam kalitesini iyileştirebilir.

Anahtar sözcükler: Erişkin bronşektazisi; pediatrik bronşektazi; cerrahi tedavi.

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Correspondence: Hakkı Ulutaş, MD. İnönü Üniversitesi Tıp Fakültesi, Turgut Özal Tıp Merkezi, Göğüs Cerrahisi Anabilim Dalı, 44000 Battalgazi, Malatya, Turkey.
Tel: +90 422 - 341 06 60 / 3907 e-mail: drhakkiulutas@yahoo.com

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Bronchiectasis is a condition characterized by irreversible abnormal dilatations of the bronchi and destruction of the bronchial wall.[1] This condition is currently recognized to be more common entity among adults than previously thought; however, it still continues to receive relatively little attention. Most studies have reported that initial symptoms of those patients can be traced back to childhood.[1] This common history reflects that childhood bronchiectasis remains an important condition, as its impact extends to the adulthood.

The optimal management for bronchiectasis still remains controversial. To date, no prospective randomized trials have been conducted to compare the efficacy of conservative and surgical treatment options.[2] It is well-known that, when preventive and medical treatment approaches for bronchiectasis fail, surgery is a major component of treatment;[3,4] however, the literature contains few data related to the results of surgical treatment for children with this condition. There is an ongoing debate regarding the relationship between surgical intervention and outcome in all age groups.

In the present study, we aimed to compare clinical manifestations, surgical indications, surgical procedures, postoperative complications, and outcomes of surgical treatment in pediatric and adult patients with bronchiectasis.

PATIENTS AND METHODS

This retrospective study included a total of 99 patients (40 males, 59 females; mean age 33.3 years; range 10 to 67 years) with bronchiectasis who underwent surgery at Turgut Ozal Medical Center, Department of Thoracic Surgery between January 2000 and December 2013 using medical records. All patients were assigned to one of two groups: pediatric patients (≤18 years) with bronchiectasis (group 1, n=28) and adult patients (>18 years) with bronchiectasis (group 2, n=71). The study was conducted in accordance with the principles of the Declaration of Helsinki.

In all cases, the initial diagnosis of bronchiectasis was based on clinical manifestations and physical examination findings. Each patient was evaluated with a chest X-ray and thoracic computed tomography (CT). A child was considered to be short stature (i.e., to exhibit growth failure) if his or her height was below the third percentile for age on a growth chart. Pulmonary function tests were performed only on the adults. All patients underwent bronchoscopy preoperatively to define the endobronchial pathology.

In most cases, surgery was performed after a trial period at least two years of conservative treatment. Of note, our center does not routinely follow patients with bronchiectasis who received medical treatment. Decisions on the need for surgical treatment are made at weekly medico-surgical meetings. The main indication for surgery was localized disease with an insufficient improvement after long-term aggressive medical management. Other indications were frequent exacerbations of the disease which hindered school attendance and adversely affected education and growth (group 1), or which interfered with normal professional or social life (group 2), or which required frequent hospitalization (either group). Persistent or severe hemoptysis, persistent abscess formation, and aspergilloma in a cystic cavity were also indications for surgery.

In the preoperative period, all patients underwent intensive chest physiotherapy and were started on prophylactic antibiotics. The surgical method was either posterolateral thoracotomy or video-assisted thoracoscopic surgery (VATS). After pulmonary resection, the bronchial stump was sutured with non-absorbable sutures or was closed using a mechanical stapler. Since we did not routinely reinforce the stump in our practice, this was only performed in patients with tuberculous bronchiectasis. In these cases, the bronchial stump was covered with a pericardial fat or peribronchial tissues. In all patients, chest physiotherapy was re-initiated soon after the operation and was continued throughout the hospitalization.

Data including age and gender of the patient, symptoms, operative procedure/s applied, postoperative morbidity and/or mortality, duration of hospital stay, follow-up duration, and surgical outcomes were recorded. In each patient, postoperative condition and general health status were also rated based on clinical examination findings in the outpatient setting, and according to the interviews with patients or their relatives. Based on the findings, the outcomes were categorized as follows: *(i)* excellent: marked improvement in social life and professional/school life after the operation, complete absence of preoperative symptoms or minimal symptoms, and does not require antibiotic therapy; *(ii)* improved: marked reduction in preoperative symptoms, but requires antibiotic therapy occasionally; or *(iii)* no change: no reduction of preoperative symptoms.

Statistical analysis

Statistical analysis was performed using the SPSS for Windows version 13.0 software (SPSS Inc., Chicago,

IL, USA). Descriptive data were expressed in mean and standard deviation (SD), and percentage. Categorical data were analyzed using the Pearson's chi-square test, and both groups were compared in terms of the ratio of patients with respective clinical features and postoperative complications. The Fisher's exact test was used to compare patient satisfaction in groups with respect to surgical procedure (i.e., complete vs incomplete resection). The Mann-Whitney U test was used to compare the length of hospital stay in the two groups. A *p* value of <0.05 was considered statistically significant.

RESULTS

Group 1 comprised 14 girls and 14 boys aged 10 to 18 years (mean: 15.5 years), and group 2 comprised 44 women and 27 men aged 19 to 67 years (mean: 40.3 years). In both groups, the most common symptom was productive cough, followed by recurrent infections. There were no significant differences in the rate of these symptoms between the groups (p=0.192 and p=0.095, respectively). Seven children (25%) showed growth failure at time of the diagnosis. In addition, hemoptysis was significantly more frequent in group 2 than group 1 (p=0.027). Table 1 summarizes the findings for symptoms in each group.

The most common cause of bronchiectasis was the lung infection. One patient in group 1 and 12 in group 2 had a history of pulmonary tuberculosis. Thirty-six adults (50.7%) suffered from recurrent lung infections since their childhood. Four patients in group 1 (14.3%) and seven patients in group 2 (9.9%) presented initially (i.e., time of original diagnosis) with middle lobe syndrome. Twelve patients in group 2 (16.9%) had idiopathic bronchiectasis. One of the adult patients had a lung abscess associated with bronchiectasis, and two had a fungus ball within a cavitary lesion.

The results for surgical procedures are summarized in Table 2. In total, 104 surgeries were performed on 99 patients. Of 29 operations in group 1, 26 were thoracotomies and three were VATS. Of 75 operations in group 2, 60 were thoracotomies and 15 were VATS. In group 1, the procedures performed were lobectomy (n=17), lobectomy + segmentectomy (n=4), bilobectomy (n=3), and segmentectomy (n=1). The corresponding numbers for group 2 were 41, 13, three, and three, respectively. Pneumonectomy was also required for two patients in group 1 and for seven patients in group 2, indicating no statistically significant difference (p=1.00).

In total, 20 patients (seven in group 1 and 13 in group 2) had bilateral bronchiectasis. Of these patients, five (one in group 1 and four in group 2) underwent bilateral complete resection of the bronchiectatic lobes. The remaining 15 patients underwent unilateral resection at the side worst affected.

Of 28 patients in group 1, 18 underwent complete resection of bronchiectatic tissue and 10 underwent incomplete resection. The corresponding numbers for group 2 were 58 and 13, respectively.

Table 1. Clinical manifestations of bronchiectasis in both groups

Symptom	Group 1 (n=28)		Group 2 (n=71)	
	n	%	n	%
Productive cough, sputum	26	92.9	70	98.6
Hemoptysis	3	10.7	23	32.4
Growth failure, sputum, cough	7	25	-	-
Recurrent infections	9	32.1	36	50.7

Table 2. Surgical procedures performed in both groups

Surgical procedure	Group 1 (n=28)		Group 2 (n=71)	
	n	%	n	%
Lobectomy	17	60.7	41	57.7
Bilobectomy	3	10.7	3	4.2
Lobectomy + segmentectomy	4	14.3	13	18.3
Segmentectomy	1	3.6	3	4.2
Pneumonectomy	2	7.1	7	9.9
Bilateral lobectomy	1	3.6	4	5.6

Table 3. Postoperative complications in both groups

Complication	Group 1 (n=29 operations)		Group 2 (n=75 operations)	
	n	%	n	%
Atelectasis	4		4	
Prolonged air leak	2		8	
Bronchopleural fistula	-		2	
Hematoma	2		-	
Lobar torsion	-		1	
Chylothorax	-		1	
Subcutaneous fat necrosis	-		1	
Cerebrovascular accident	-		1	
Total	8	27.6	18	24

In all 99 patients, surgical specimens were examined by a single pathologist who confirmed the diagnosis. Histologically, all specimens showed varying degrees of bronchiectatic changes, including destruction of the lung parenchyma, markedly enlarged bronchial circulation, and frequent bronchopulmonary anastomosis. However, no active tuberculosis was histopathologically shown in the surgical specimens of the cases of tuberculous bronchiectasis.

Eight operations (27.6%) in group 1 and 18 operations (24%) in group 2 were associated with postoperative complications (p=0.814) (Table 3). There was no intra- or postoperative mortality. The mean length of hospital stay was not significantly different between the groups (10.8±6.4 days for group 1 vs 9.9±5.4 days for group 2). The median duration of hospitalization was nine days (range: 4 to 30 days) in group 1 and eight days (range: 4 to 27 days) in group 2.

The median follow-up times were 75 months (range: 18 to 134 months) for group 1 and 57 months (range: 4 to 146 months) for group 2. The outcomes were rated excellent (asymptomatic) for 23 patients in group 1 and 63 patients group 2, and improved (positive changes in the preoperative symptoms) for five patients in group 1 and seven patients in group 2. All of the pediatric patients showed some degree of improvement, while only one of the adult patients showed no improvement. All seven children who showed growth failure at time of the diagnosis attained their expected growth potential after surgery. In group 1, there were no significant differences in the patient satisfaction (i.e., ratio of symptomatic vs asymptomatic patients) between the subgroups those underwent complete resection and incomplete resection (p=0.344). However, in group 2, there was a significant difference in the patient satisfaction between these subgroups (p=0.033), and overall patient satisfaction was significantly different between group 1 and group 2 (p=0.002).

DISCUSSION

Although the current prevalence of bronchiectasis worldwide is not definitively known, previous studies have suggested that the prevalence may have decreased in recent years due to childhood immunizations and greater use of broad-spectrum antibiotics, particularly in developed countries.[1,5] However, bronchiectasis is currently being diagnosed more frequently throughout the world. One recent study from the United States documented a significant rise in the prevalence of this condition.[6] Many reports also suggest that bronchiectasis is now recognized as an important and common cause of respiratory disease, and that it remains a potentially serious condition worldwide.[7-10]

Bronchiectasis should be suspected in any individual who presents with persistent daily cough and mucopurulent sputum.[11] In addition to chronic productive cough, the most common symptoms are recurrent upper and lower respiratory tract infection, and hemoptysis.[11] With the exception of hemoptysis, which is rare in childhood bronchiectasis, there is no clear distinctive manifestation between pediatric and adult cases. Twenty-five percent of our pediatric patients showed growth failure as a prominent symptom of bronchiectasis. In the literature, it has been reported that, in 21 to 80% of cases, symptoms of bronchiectasis begin in childhood.[12-14] Among our 71 patients, this rate was 50.7%, indicating the importance of treating bronchiectasis effectively in childhood.

Chest radiography can help to identify bronchiectasis, but it is not diagnostic. High-resolution CT has been

reported to be the gold standard method in the diagnosis of this condition;[11] however, most descriptions of the radiological criteria for diagnosis pertain only to adults. Gaillard et al.[15] reported that these criteria were not validated in children and that this could lead to overdiagnosis of the condition. Furthermore, bronchiectasis in childhood may progress to become irreversible by extending to new sites and evolving into saccular changes;[16] however, in contrast to adults, affected children may exhibit radiological resolution or significant improvement in bronchial dilatation over time.[13,15] Gaillard et al.[15] also achieved complete resolution in six of 22 children with bronchiectasis, and Eastham et al.[13] achieved complete resolution in six of 18 children. Both authors suggests that children should only be diagnosed with established bronchiectasis, if they exhibit *(i)* a combination of persistent clinical signs over a two-year period, and *(ii)* irreversible change on an interval CT scan during this period.[13,15] In our series, except some patients with symptomatic destroyed lung or lobe, decision on the need for surgical treatment was made at medico-surgical meetings after a medical therapy period of at least two years.

Irrespective of age, management of these patients requires a comprehensive multi-modal therapeutic approach which encompasses surgical intervention. Aggressive medical therapy is recommended before surgical decision. However, in any circumstances such as uncontrolled disease and/or acute pulmonary infection, bronchiectasis may spread from an affected area to adjacent normal tissues with direct spread of infection and inflammation which ultimately results in reduced lung functions (i.e., reduced diffusion capacity), and increased airway hyper-reactivity.[17] Andrade et al.[18] suggested that, in many cases, optimal clinical control of disease was unable to be achieved due to low socioeconomic status of patients. We believe that uncontrolled disease affects not only pulmonary function, but also the quality of life of the patient. The insistence on medical therapy for patients with localized disease is also associated with higher disease burden (i.e., increased morbidity, reduced quality of life, and increased socioeconomic cost of long-term management). Difficulties in relieving symptoms or complications through a medical approach lead to consideration of surgery.

Postoperative quality of life is one of the critical factors in deciding surgical management of bronchiectasis. The principal goal of surgical resection in such cases is to eradicate the diseased portions of the lung, while preserving the healthy lung parenchyma as much as possible.[19] The rate of complete cure in patients undergoing complete resection has been reported between 65 and 85%.[3,20-22] The complete cure rate in patients undergoing incomplete resection is 9 to 43%.[21-23] As in other series, we found that patients in both age groups who underwent complete resection had improved prognosis than those who underwent incomplete resection.[12,22,23] In reported series, incomplete resection rates range from 9.3 to 43.5%.[20,23-25] Many authors have claimed that, in selected cases, non-localized bronchiectasis should not be regarded as a contraindication to surgical therapy and even incomplete resection often provides considerable benefit to the patient who has non-localized, but mainly resectable disease.[18,20,23,26] Agasthian et al.[3] reported that 25 of 26 patients with bilateral bronchiectasis had incomplete resection. In this group that had an incomplete resection, 21.4% of cases were asymptomatic, 50% of cases were improved, and 28.6% of cases showed no improvement. Other authors reported that, of those patients who underwent incomplete resection, 56 to 86% benefited (cure or improvement) from surgery.[21-23,25] In our study, we also observed that incomplete resection led to substantial improvement in symptoms by converting severe bronchiectasis to mild disease, and this was particularly notable in children. However, the criteria for resection must be more selective for patients who are candidate for incomplete surgery.

In the patients with bilateral bronchiectasis, in accordance with the findings of Sanderson et al.,[26] our approach for this patient group is to operate on the worst-affected side first. We found that unilateral resection was satisfactory in most such cases and resection of bronchiectasis in the contralateral lung could be usually safely postponed, until an older age in children. In their series, Prieto et al.[23] showed, of 10 patients with bilateral bronchiectasis, only two underwent second operation for contralateral lung. The authors highlighted that one of the reason of this decision was due to the clinical improvement after the initial procedure.[23] In cases where bronchiectasis has totally destroyed the lung parenchyma, pneumonectomy has proven to be the most effective way to manage related complications.[23,27] Pneumonectomy can be performed in children and yield acceptable rates of mortality and morbidity,[27] and this procedure can achieve satisfactory long-term survival in these patients.[27,28] Based on a 30-year observational study including 230 patients aged 2 to 40 years who underwent pneumonectomy demonstrated that, compared to the adults, children were able to tolerate pneumonectomy well and experienced less functional disability after this

operation.[29] Similarly, in our study, pneumonectomy was performed with satisfactory outcomes in two children.

In the literature, the complication rates for surgical treatment of bronchiectasis in all age groups range from 8.8 to 33.4%.[3,4,12,22-24,30-32] Some authors have also reported morbidity rates ranging between 7.4 and 36% for pediatric patients.[18,25,33,34] In our series, we observed nearly equal morbidity rates for our adult and pediatric patients who were treated surgically (27.6% and 25.3%, respectively; p>0.05), and most complications were minor, such as transient atelectasis or prolonged air leakage. The mortality rates after surgical treatment for bronchiectasis in adults are relatively low, ranging from 0 to 2.2%,[3,4,12,22-24,30-32] while these rates are between 0 and 5.6% in pediatric cases.[18,25,33,34] In our study, there was no intra- or postoperative mortality in either group.

Earlier, patients with bronchiectasis were treated surgically via thoracotomy. Currently, however, VATS resection is an established alternative approach to conventional open surgery for selected patients.[35-37] In patients with bronchiectasis, surgical intervention via the thoracoscopic approach is challenging due to the presence of vascular pleural adhesions and bronchial artery hypertrophy. However, several recent studies have demonstrated the technical feasibility of VATS resection for bronchiectasis.[35-38] Lower morbidity, improved functional status, and shorter hospital stays have been reported for patients undergoing these minimally invasive procedures.[35,36] In the present study, we used VATS successfully in 18 patients in our series. In the future, we believe that more experience with VATS would allow surgeons at our center and elsewhere to perform more complicated procedures. More experience and success with this technique can lead to earlier referral for surgical resection in cases of bronchiectasis.

In conclusion, surgical treatment is a safe procedure for children with well-localized bronchiectasis. Similar to adults, surgical management can be performed with non-life-threatening morbidity and a high expectancy for cure. Surgical management should be considered as an important component in the management of selected children with non-well-localized bronchiectasis, bilateral disease, or where known residual disease is left after resection, by a multidisciplinary team. Resecting the most affected tissue offers the possibility of significantly reduction of symptoms and can improve the quality of life in these cases.

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