# The Surgical Management of Childhood Bronchiectasis

## A Review of 96 Consecutive Pulmonary Resections in Children with Nontuberculous Bronchiectasis

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**In 195 children with nontuberculous bronchiectasis, periodic bronchography and clinical examinations were conducted over a period of 16 years (average 9.4 years). This has provided a critical assessment of surgical accomplishments in 96 consecutive resections and a parallel observation of 111 cases not submitted to resection. The final clinical assessment of the surgical cases shows 75% to be well or much improved, 22% to be improved, and 4% unchanged, while patients not submitted to resection have remained largely unchanged (69%) or have become worse (23%). The isolated superior segment can be preserved in children with good results, provided there is clear bronchographic evidence that *the segment is entirely free of disease.* When partially diseased segments are retained and required to fill a large volume, there is a tendency for *even slightly altered bronchi to deteriorate postoperatively.* Serial bronchography has proved helpful in determining when the disease has reached a mature, stable state and in planning the extent of resection.**

THE APPROPRIATE SURGICAL MANAGEMENT of childhood bronchiectasis is a difficult problem for which the surgeon finds few useful guidelines in the recent North American literature. Thirty years ago, surgery for bronchiectasis was common, the modern techniques of segmental pulmonary resection having been largely developed for the management of this disease. Today, childhood bronchiectasis is seldom diagnosed even in major medical centers, and many well-trained thoracic surgeons have had little or no experience with its management. Recently, bronchiectasis has been most commonly reported from Africa,[1] from India,[2] and among the aborigines of Australia,[3] and New Zealand.[4]

The high incidence of bronchiectasis in Alaska Native children has been reported previously.[5] This paper is a review of a 24-year experience with the surgical management of childhood bronchiectasis in this population.

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## Materials and Methods

### *Background*

In about 1964 it was recognized that the high incidence of childhood bronchiectasis occurring in a small population served by a single referral center offered unique opportunities for study. Accordingly, cases of bronchiectasis were identified and followed by periodic clinical and bronchographic evaluations. Limitations imposed by restricted resources usually required that these evaluations be carried out in small outlying Public Health Service field hospitals, but a simplified, safe technique for bronchography in children performed under topical anesthesia permitted serial bronchographic studies in a high percentage of cases.[6] Bronchography was performed two or more times in 78%, and three or more times in 54%. When seen by a physician, either in the village, the field hospital, or at the referral center, each child was evaluated for specific symptoms and for general health status (as described below). These procedures have been followed over a 16-year-period through 1980.

### *Clinical Material*

For a patient to be included in the study, bronchographic evidence of bronchiectasis before the age of 17 years was required. The earliest diagnosis was made at the age of 5 months, and 77% were diagnosed before the age of 11 years. The mean age at diagnosis was 7.7 years. The male/female ratio was 1/1.

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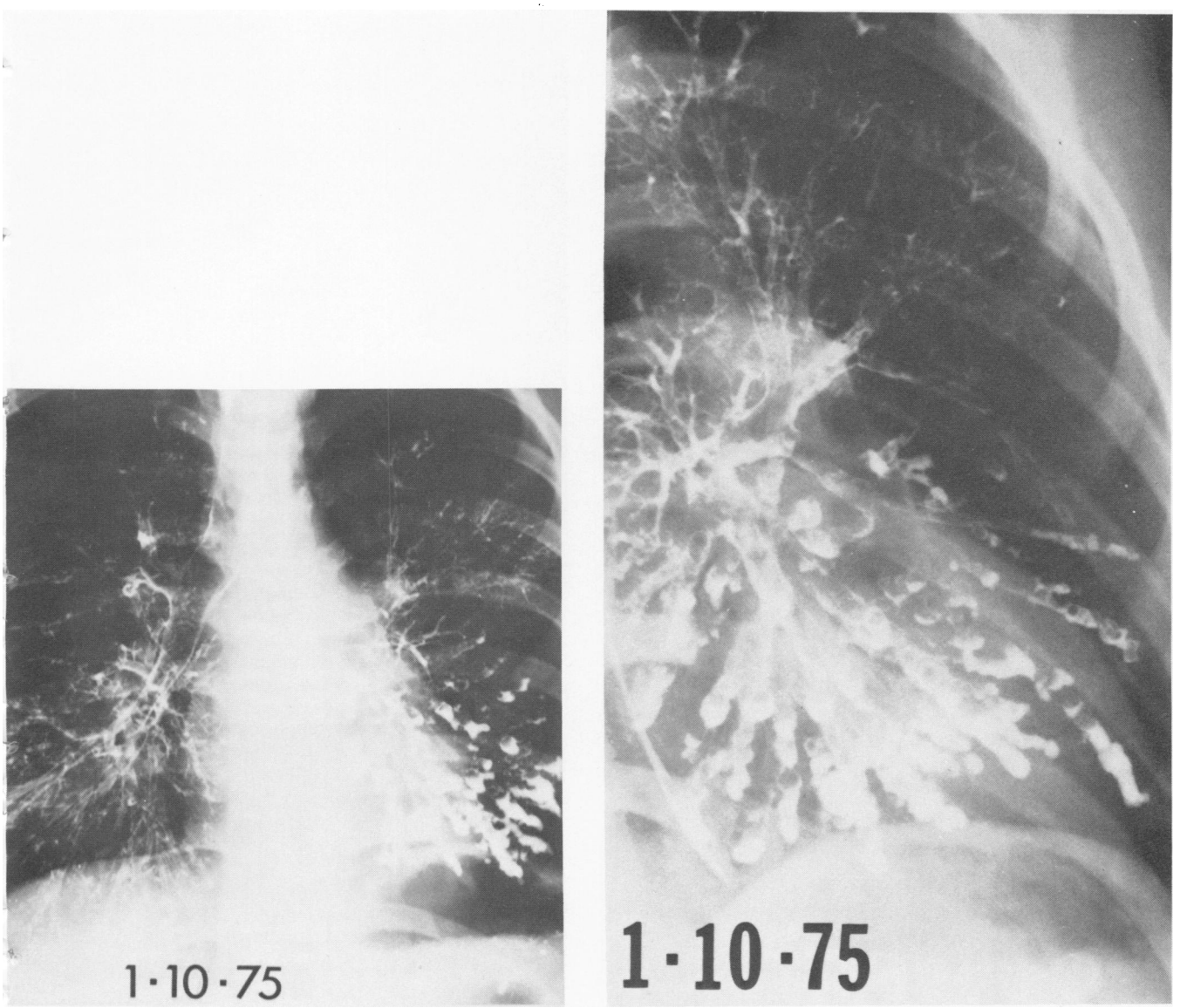


FIG. 1A. Two views of the preoperative bronchograms of a 16-year-old female who had been followed for seven years with periodic bronchography.

There were 250 cases of childhood bronchiectasis diagnosed between 1956 and 1980. They come from a population of approximately 80,000 Alaska Native Eskimos, Indians, and Aleuts. Of these, 118 patients underwent 136 resections. There were 37 patients classified as having tuberculous bronchiectasis, seven patients had surgery performed elsewhere, and in ten individuals there was unequivocal evidence of bronchiectasis on chest films prior to the age of 17, but their first bronchogram was not obtained until after this age. All of these cases (54) will be excluded from further review. An analysis of the remaining 195 children, requiring 96 consecutive pulmonary resections in 84 patients, forms the basis of this paper.

*Preoperative Surgical Objectives*

Many factors must be considered in selecting a patient for surgery. Each case is, in practice, decided on its own merits. However, for the purpose of this review, cases have been divided according to one of three preoperative objectives.

1. Curative resections: Patients in this category generally have well-localized, unilateral disease causing significant symptoms; resection of all known disease in these patients can be anticipated with a single procedure. The objective is to eliminate all symptoms. An example of a case in this category is illustrated in Fig. 1.


FIG. 1B. Because of persistent, troublesome purulent sputum production, a "curative" resection of the left lower lobe and the lingula segments was carried out January 15, 1975. Note the good volume of the remaining two segments in the postoperative bronchogram. The functional value of these segments was demonstrated by recent lung scan, showing that 25% of overall pulmonary blood flow was carried by these two residual segments. She has been symptom-free for six years.

2. Intermediate resections: These patients have less well-localized disease; most of the disease can be resected without great risk. Patients with bilateral but well-localized disease are also included in this group. The objective is to reduce symptoms significantly (Fig. 2).
3. Salvage resections: Occasionally, patients with severe, bilateral, and often poorly localized disease are submitted to resection in a desperate attempt

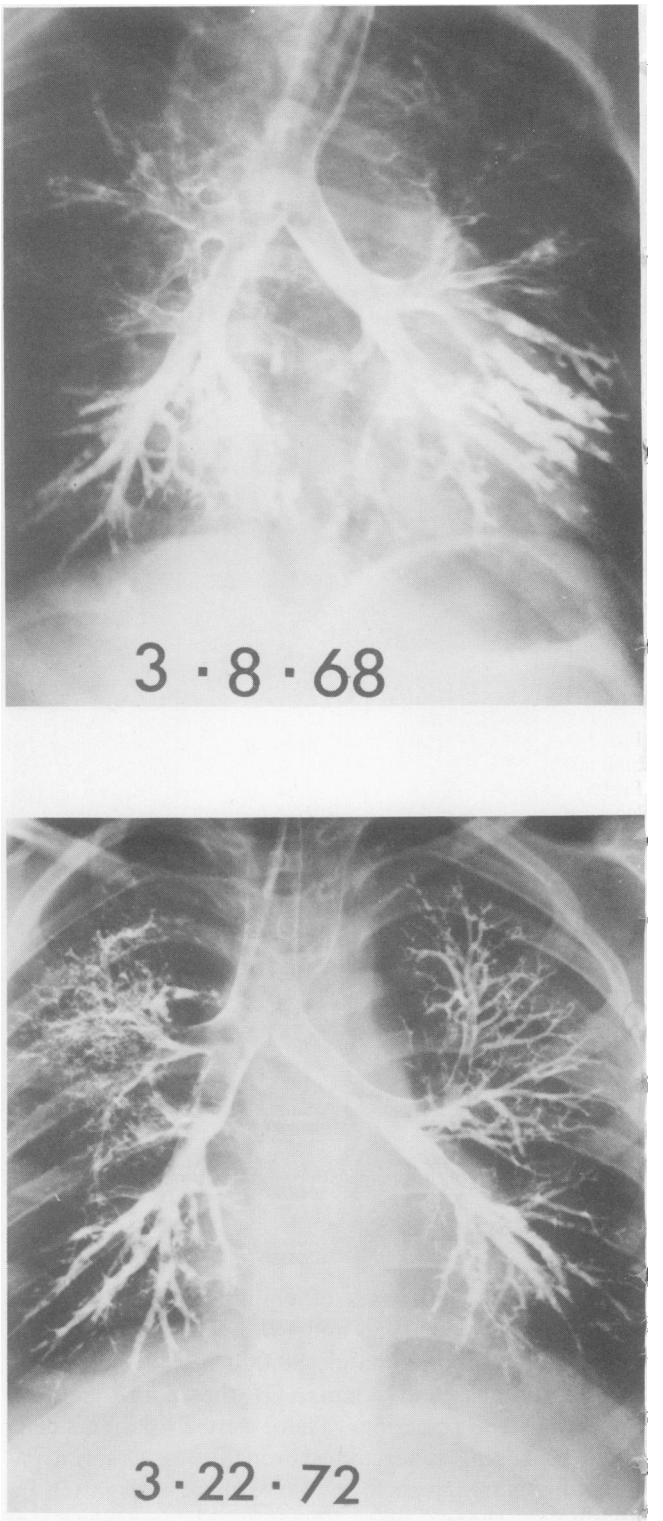


FIG. 2. An "intermediate" resection is illustrated by this case in which bilateral resections for fairly well-localized disease were required. A. Bronchograms on March 8, 1978 demonstrate disease of the RML and the lingula division of the LUL. Resections of the RML and lingula were carried out at age 8 years. B. Some known residual RUL disease seen on the bronchograms of March 22, 1972 accounts for minimal sputum production at the last clinical assessment nine years after operation.

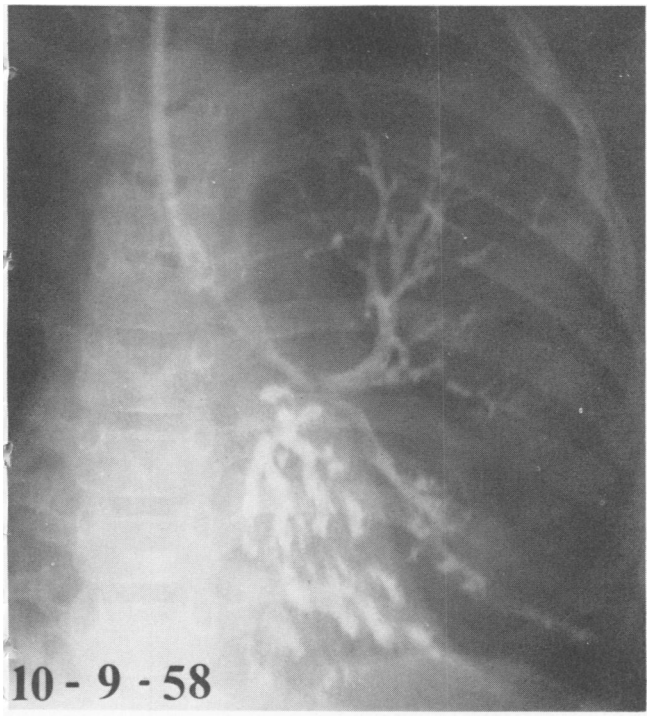


FIG. 3A. A "salvage" resection is illustrated by a case of far-advanced bilateral bronchiectasis diagnosed at 3 years of age (October 9, 1958). Profuse sputum production and failure to thrive necessitated prolonged hospitalization.

to reduce disabling symptoms to the point that they can be controlled by intensive medical management. It is anticipated that the resection is a high-risk procedure. The objective is to control disabling or life-threatening symptoms (Fig. 3).

*Postoperative Results*

Postoperative results were classified into four descriptive categories. The definition of each follows.

1a. Well: Patients in this group have no significant symptoms and no residual disease on bronchography. Occasional sputum production with lower respiratory tract infections is accepted.
1b. Much improved: Patients in this group show a pronounced reduction in symptoms, but with some residual disease on bronchograms, or mild clinical symptoms.
2. Improved: Symptoms are still significant and the patient's daily life may be affected, but symptoms are reduced in degree.
3. Unchanged: The patients' symptoms persist, unchanged.
4. Worse: Symptoms are more severe than at the time of preoperative assessment.

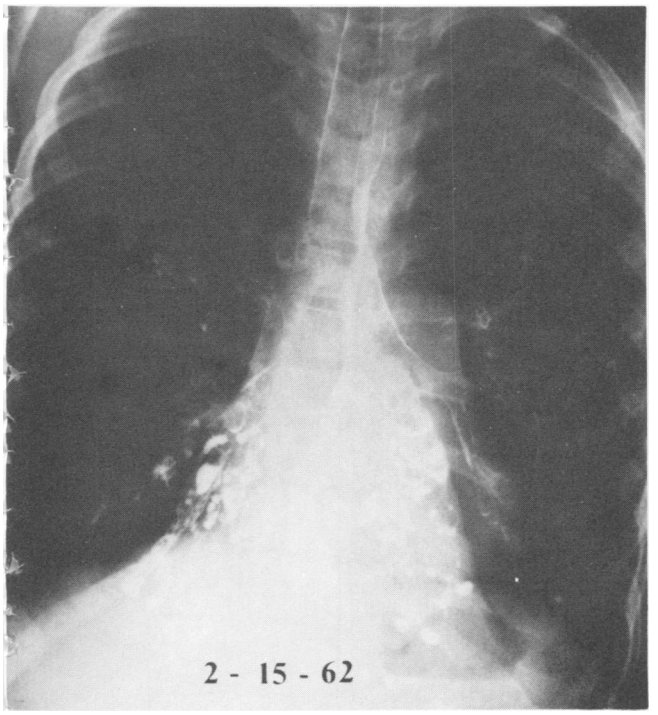


FIG. 3B. The preoperative bronchograms prior to uneventful bilateral resections were obtained at age 7 years, with the RLL and subsegment of the RUL resected February 22, 1962, and the LLL and inferior lingula segment resected March 29, 1962.

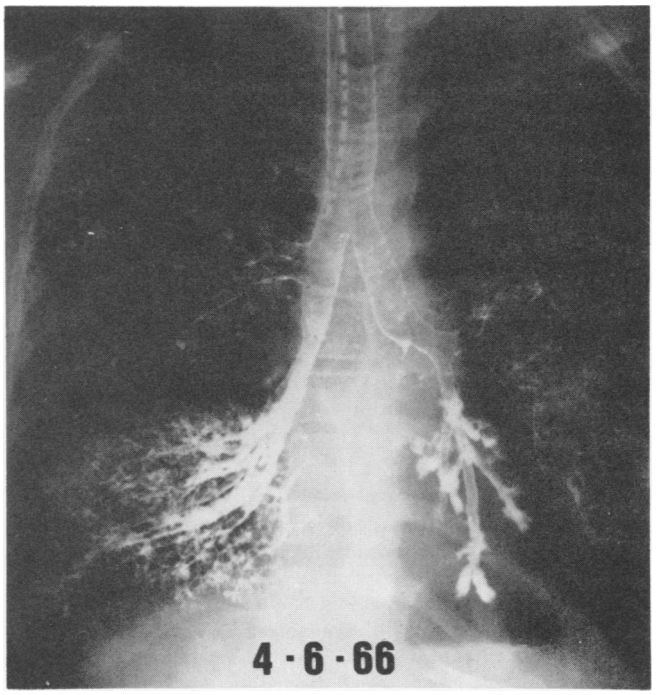


FIG. 3C. Note the postoperative deterioration or progression of disease in the remaining superior lingular segment of the LUL. The patient is now 26 years old, has a successful marriage with two children, and has minimal to no pulmonary symptoms.

*Surgical Procedures*

Preoperative preparation usually consists of intensive respiratory therapy and postural drainage. Immediate preoperative and postoperative antibiotics are routinely used.

Surgery is carried out in the lateral position, entering the chest through the fourth or fifth interspace. Every effort is made to keep the operative lung well expanded during the resection. High volume and high negative pressure pleural suction (−50 to −60 cm $H_2O$) is used postoperatively on all thoracotomies and is believed to be of great importance. Early ambulation is encouraged, and very close postoperative surveillance is mandatory. Retained secretions are frequently a problem, especially in children with known residual disease. Although postural drainage and respiratory therapy are routinely used, transnasal, endotracheal suction has been the most effective means of removing secretions. A few cases, especially those with bilateral disease or those with a retained, isolated superior segment, required frequent endotracheal suction,* sometimes as often as every one to three hours. Bronchoscopic aspiration of secretions has been employed only occasionally.

## Results

Of the 84 surgical patients with nontuberculous bronchiectasis, postresection bronchography has been obtained in 82%. Postoperative clinical evaluations were made in all cases. The average period of postoperative clinical follow-up has been 10.1 years (range 2–247 mo.). The final results are presented by surgical indication or objective, and by the extent of resection (Table 1 and Fig. 4).

It is apparent that when virtually all disease can be resected by one operation, the results have been almost uniformly excellent. The results in the intermediate and salvage groups were also quite good. Seventy-four per cent of all patients were considered well or much improved, 22% were improved, and 4% remained unchanged. None was made worse by the surgical treatment. Sixty-seven per cent of the resected cases are currently asymptomatic.†

TABLE 1. *Surgical Results by Indication and Extent of Resection*

| Extent of Resection | Curative | | | | | Intermediate | | | | | Salvage | | | | | All Cases | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | No. of Patients | Well | Much Improved | Improved | Unchanged | No. of Patients | Well | Much Improved | Improved | Unchanged | No. of Patients | Well | Much Improved | Improved | Unchanged | No. of Patients | Well | Much Improved | Improved | Unchanged |
| Lobe or less | 17 | 16 | 1 | 0 | 0 | 15 | 0 | 9 | 6 | 0 | 0 | 0 | 0 | 0 | 0 | 32 | 16 | 10 | 6 | 0 |
| More than lobe to less than lung | 16 | 11 | 4 | 1 | 0 | 12 | 0 | 6 | 6 | 0 | 5 | 0 | 1 | 4 | 0 | 31 | 10 | 10 | 11 | 0 |
| Lung | 5 | 5 | 0 | 0 | 0 | 2 | 0 | 2 | 0 | 0 | 6 | 0 | 2 | 1 | 3 | 13 | 5 | 4 | 1 | 3 |
| Bilateral resection | 0 | 0 | 0 | 0 | 0 | 4 | 1 | 3 | 0 | 0 | 2 | 0 | 2 | 0 | 0 | 6 | 1 | 5 | 0 | 0 |
| Total cases | 38 | 32 | 5 | 1 | 0 | 33 | 1 | 20 | 12 | 0 | 13 | 0 | 5 | 5 | 3 | 84 | 33 | 30 | 18 | 3 |
| Per cent | 100 | 84 | 13 | 3 | 0 | 100 | 3 | 60 | 36 | 0 | 100 | 0 | 38.5 | 38.5 | 23 | 100 | 39 | 36 | 22 | 4 |

* The technique used is basically the same as that described for bronchography[16] but is performed without topical anesthesia.

† By definition, patients classified as well have no residual disease on their postresection bronchograms. Not infrequently, patients have some residual disease but are asymptomatic. Four patients in the curative group, ten patients in the intermediate group, and two patients in the salvage group, though having some residual disease on their postoperative bronchograms, are currently asymptomatic and were classified as much improved. Consequently, while only 39% were classified as well, there are actually 67% who remain asymptomatic following resection.

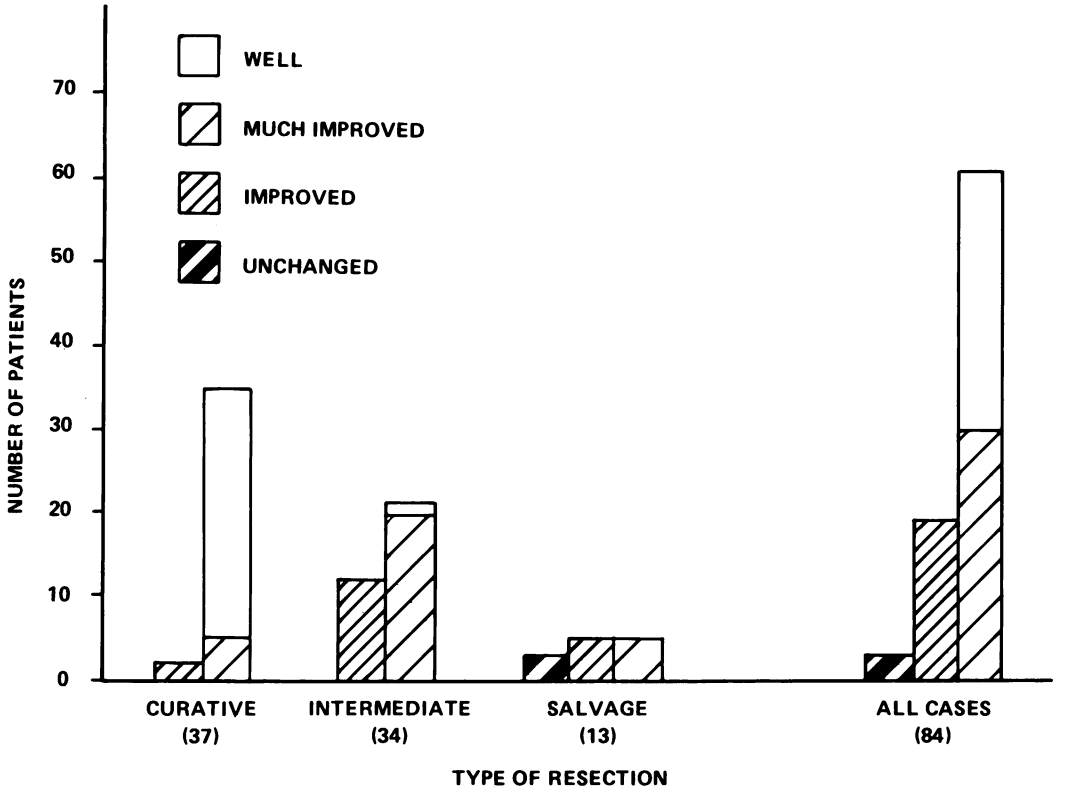


FIG. 4. A clinical assessment of postoperative results by surgical objective is illustrated. When disease was localized and the objective was a *curative* resection, results were uniformly good. Although the results were not as good when the operation was undertaken as a salvage procedure, 77% of the patients were improved or much improved.

Bilateral disease was observed in 85 patients (44%), and 31 of these underwent resection, including six children who had bilateral resections. In six cases a bilateral resection was initially planned, but sufficient improvement resulted from the initial resection that the second procedure has not, as yet, been required. Of the 83 patients selected for surgical treatment, two cases required a second resection to control symptoms from residual segments unrecognized as being diseased at the first operation.

TABLE 2. *Surgical Complications*

| Complications | Treatment | Number of Patients | Final Clinical Results |
|---|---|---|---|
| Bronchopleural fistula after resection of entire left lung except for superior segment, left lower lobe | Phenumonectomy | 1 | Improved |
| Atelectasis of superior segment, persistent | Resection of superior segment two years later. | 1 | Much improved |
| Atelectasis of superior segment, persistent | None | 1 | Eventual reexpansion Improved |
| Respiratory insufficiency, postoperative to salvage pneumonectomy | Mechanical ventilation for four days. | 1 | Unknown* |
| Postoperative pneumothorax, postoperative days six and ten | Tube drainage | 2 | Prompt resolution No long-term affect |
| Apical air space | Tube drainage | 2 | " " |
| Chylothorax | Tube drainage | 1 | " " |
| Total | | 9 | |

* Long-term clinical assessment was not possible because patient died four months postoperatively of causes unrelated to her pulmonary disease.

*Surgical Complications*

Surgical complications were infrequent (Table 2). Only four had any long-term adverse effect. Three of these occurred following resections that resulted in retained superior segments: one had a bronchopleural fistula requiring completion of the pneumonectomy, and two had persistent atelectasis of the superior segment. The fourth long-term complication resulted when massive pulmonary sepsis extended to the remaining lung following a salvage pneumonectomy. The patient developed severe respiratory insufficiency, requiring assisted ventilation for four days. The remaining complications, all short-term, responded to tube drainage without sequelae. There were no complications among the six children requiring bilateral resection and only one among the 14 pneumonectomies.

*Special Surgical Problems*

*Isolated superior segment.* The question of whether the superior segment should be preserved when the major disease is limited to the basal segments is not easily answered in children. The superior segment, even when moderately diseased bronchographically, will usually present with a large volume and a normal appearance at surgery. It is a very valuable space filler, and there is often a strong temptation to preserve it. In 12 cases, this segment was preserved. Detailed study of these cases reveals that in eight (66%), some clear-cut though often minimal disease could be identified in this segment preoperatively. In five of the eight, some postoperative deterioration of this segment occurred. Two of the latter required resection of the retained superior segment. The complication rate in these patients was high, with three of the four most serious complications of the entire series coming from this group of eight patients. On the other hand, eight of the 12 currently have either minimal or no symptoms. In five cases the preoperative bronchogram showed no evidence of disease in the superior segment. Postoperative bronchograms, obtained in four of the five cases, have been normal.

*Splitting the basal segment.* Most surgical textbooks state that the basal segments of the lower lobe should not be surgically subdivided. In our efforts to preserve as much pulmonary function as possible, we have, in ten cases, split one or two segments from the basal group in order to remove full-blown but isolated disease. There were no complications in this group. The diseased segment is usually identified at surgery by a small area of palpable bronchiectatic sacs or a small wedge of atelectasis. Beginning with this often small area, a retrograde open-wedge dissection is carried out, tracing the disease to its bronchus. The bronchus is traced centrally until the appropriate segmental level is reached. The segment is then isolated, and only vessels to the diseased segment are ligated. This retrograde resection technique has not been difficult, and the results of these resections have been quite good. We recommend this technique as do Shaw and Logan.[15]

*Net Experience*

In substance, the net experience with resection for bronchiectasis is encouraging, and it is our feeling that resection can be offered with little morbidity, almost no mortality, and with a high expectation of amelioration of symptoms. We cannot, as of yet, know the long-term outcome for these patients, but in follow-up into young adulthood, we see few signs of deterioration. We do feel that there are at this time, perhaps, somewhat fewer indications for surgery in even gross bronchial dilations than there are in symptomatology. A proper evaluation of what may be expected is, of course, vital to this consideration and abrings us to our nonsurgical experience with this group of patients.

*Nonsurgical Experience*

Since there has been an encompassing enthusiasm for treating most suppurative pulmonary disease by nonsurgical means in the antibiotic era, it has become extremely important to document the clinical and pathological course of bronchiectasis that has not been subjected to surgical intervention. It was with this in mind that the present study was directed, early in the 1960s, toward close attention to bronchographic follow-up and clinical evaluations of a large group of nonresected patients.

Surgical indications have become increasingly restrictive during the period of this study, 1956 to 1980. This is not because the results of surgery have been discouraging; on the contrary, they have been excellent. Rather, this occurred because of an increasing concern to assure that the results of conservative management could be carefully compared with those treated surgically. Very little has occurred so far to make us regret this degree of procrastination.

There are 111 patients who have not had surgery. Thirty-eight are not surgical candidates: 17 have far advanced, bilateral, highly symptomatic, panbronchiectasis; five have less severe but poorly localized disease; 11 have minimal disease with no indication for surgical intervention; and five are not considered surgical candidates for other medical reasons. However, there are 73 patients who have localized disease and are considered potential surgical candidates. In 30, the disease is limited to one lobe (RUL in 15, RML in four, and a lower lobe in 11), with five having moderate or severe

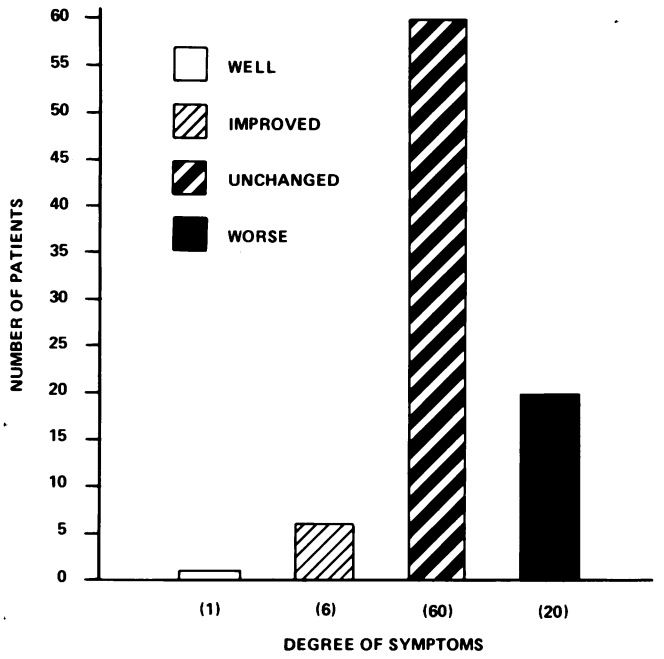


FIG. 5. Results of nonoperative treatment in 87 surgical candidates with prolonged observation (average 7.3 years), 28 of whom subsequently had surgery. Numbers in parentheses represent the actual number of patients in each category.

symptoms and 25 having minimal or no symptoms. Some of the patients have far-advanced disease but present with only minimal symptoms. All 111 patients are being monitored and will be reevaluated for surgery if their symptoms become more severe, or if they become less responsive to medical management. The primary physician is usually involved in the assessment and treatment of these patients and in the decision for surgical intervention. During recent years nearly all patients selected for surgery (about 15) were referred in this way by the primary physician. Currently, 52 of the 73 patients (71%) have minimal or no symptoms, 17 (23%) have moderate symptoms, and in four (5%) the degree of symptomatology is classified as severe.

An assessment of the clinical status of 87 surgical candidates who had prolonged observation (average 7.3 years, range 2 to 11 years) is presented in Fig. 5. Fifty-nine of these are from the nonresected group, while 28 are from the resected cases with their assessment made prior to resection. Patients not considered surgical candidates for the previously mentioned reasons are excluded from this study. This is done with the objective of making these cases more comparable to those analyzed in Fig. 4. However, important differences are recognized, and the one group should not be considered a control for the second.

It is readily seen that without surgical intervention, symptoms in children with bronchiectasis remain largely unchanged or become progressively more severe. In these patients, the major symptoms are troublesome cough with persistent purulent sputum.

Without randomized study of surgical vs. nonsurgical cases, a true comparison between operative and nonoperative cases is impossible. However, the selection of those patients with serial bronchography who have been kept under observation for a period of time and then submitted to resection utilizes the patients themselves, to some measure, as their own controls. In 30 cases meeting these criteria, the degree of symptomatology was averaged at three stages during their treatment: at the time of diagnosis, immediately prior to resection, and at the last postoperative follow-up examination (Fig. 6). The mean period of preoperative observation

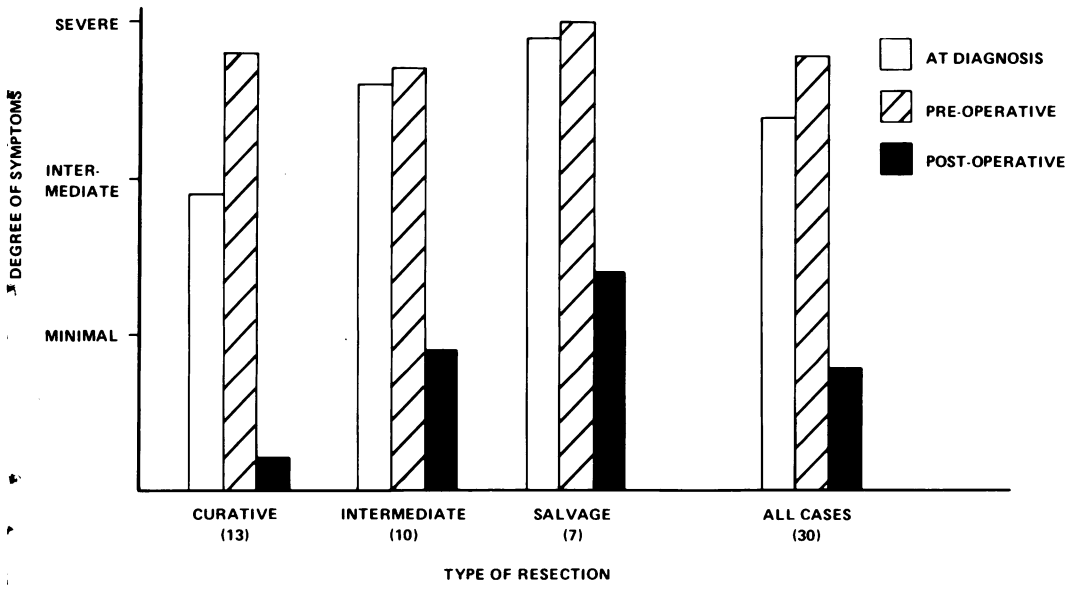


FIG. 6. The preresection and postresection symptomology, expressed in the average degree of symptoms, is compared by category of resection in 30 patients with prolonged preoperative evaluations. The mean period of preoperative observation was 4.8 years (range 2–12 years), and 7.7 years for the mean period of postoperative observation. The three degrees of symptoms have arbitrarily been assigned numerical values; i.e., 1 for minimal symptoms, 2 for moderate symptoms, and 3 for severe symptoms. The values so assigned have been averaged for each category of resection.

was 4.8 years, and the period of postoperative clinical follow-up has been 7.7 years. A significant reduction in the degree of symptoms is observed following resection. With few exceptions, neither the nonoperative nor the postoperative patients in this experience have received intensive medical management due to the patients' isolation from medical care facilities.

## Discussion

There is virtually no pulmonary function or pulmonary arterial blood flow through a lobe or lung that is totally destroyed by bronchiectasis. This has been described by Liebow, et al.[7] We have confirmed this through bronchospirometric and lung scan studies. As a consequence, resection of a destroyed bronchiectatic lobe or lung has little effect on pulmonary function. On the other hand, when a segment or a lobe is only partially diseased, there may be important residual function in the diseased segment. As a result, the choice of whether to remove or preserve the area is often more difficult. Removal of such a segment sacrifices both function and vascular bed.

Our policy has been to be as conservative of pulmonary tissue as possible. Early in this experience, we were more concerned that localized disease would spread to uninvolved areas of the lung. Consequently, we often resected at an early age and more frequently preserved segments that were partially diseased. Observations on the natural course of bronchiectasis have shown that *progression* of disease from early, often ill-defined changes to full-blown bronchiectasis occurs commonly in young children (approximately 26% in this series), while *extension* to healthy, previously undiseased tissue is rare (about 2%). Currently, we are more inclined to delay surgery until the full extent of the disease has declared itself (usually between the ages of 6 and 12 years). We also are inclined to sacrifice isolated areas such as the superior segment or lingula if there is bronchographic evidence of even minimal disease. Having several sets of bronchograms has proven helpful in making appropriate decisions.

There is ample evidence in this material that isolated basal segments or lingular and middle lobe segments may be resected without significantly increased technical challenge. Most residual situations that leave, for example, only the apical posterior and anterior segments on the left to fill a large space will work out well, both bronchographically and in so far as filling space is concerned. The functional value of these retained segments has previously not been well documented. In this study six cases had only the upper division of the left upper lobe retained on the left, and in eight cases only the right upper lobe was retained on the right. Recently, six patients followed 10–12 years postoperatively underwent differential pulmonary function studies, including lung scans and bronchospirometry, to determine the function of these retained segments. In the three patients that had only the two segments of the upper division of the left upper lobe retained, the average function was about 28% of overall function (45% is normal for the entire left lung). In those following resections of the right middle and lower lobes, the function of the residual three segments was about 35% (55% is normal for the right lung). (Wilson—unpublished data). We have come to recognize that bronchi slightly altered in preoperative bronchograms in segments remaining after resection and asked to overexpand to fill space or to shift to a more dependent position present rather ominous possibilities. Such previously altered and slightly distorted bronchi *may be expected to progress* torward significant bronchiectasis in many if not most instances. This is certainly a factor to be considered in decisions to resect under less than ideal indications.‡

### *Surgical Indications*

We list our current indications for resection with the understanding that they are not definite recommendations, but are a necessary phase of the evaluation process used in this study.

*Clear-cut indications.*

1. Localized disease producing severe symptoms: a) profuse sputum; b) fetid breath; c) severe cough or other symptoms that interfere with a normal life pattern.
2. Threatening hemorrhage from a demonstrated focal, segmental, or lobar source.
3. Resectable disease of significant severity associated with failure to thrive.
4. Resectable disease of a demonstrated site of recurrent, acute lower respiratory infections.

*Less clear-cut indications.*

1. Evidence of unstable disease associated with significant progression or extension of resectable disease.
2. Bronchiectasis not easily or totally resectable but associated with a failure to thrive.
3. Bronchiectasis not easily or totally resectable associated with life-threatening or truly disabling symptoms: a) profuse sputum; b) fetid breath; c) hemorrhage; d) severe recurrent focal infection episodes.

‡ In four cases, progression of known residual disease has necessitated a second resection before a good result was finally obtained.

4. Localized disease producing minimal to moderate symptoms.

Of the few studies in the English language dealing with the surgical management of childhood bronchiectasis, most have results quite similar to those presented in this study.[2,3,8-16]

Shaw and Logan described their experiences with 218 patients in Ireland over an 11-year period, all of whom were resected. Eight-nine per cent had nontuberculous bronchiectasis and 47% were resected before the age of 20 years.[16] Considering medical treatment the preserve of hypochondriacs, their policy was to operate on all symptomatic cases in which resection would not produce respiratory impairment. Unilateral resections in 193 patients "cured" 52% and "improved" another 43%. Bilateral resections in 25 patients "cured" only 16%, but 24% were considered "satisfactory" and 60% were "improved."

Sanderson et al. reviewed 393 consecutive cases of bronchiectasis followed for 1–15 years. About two thirds were managed surgically, and one third conservatively. It was their conclusion that the current bias toward conservative management was unjustified.[15]

## Conclusions

From a review of 195 cases of childhood bronchiectasis followed over a 16-year period by serial bronchography, several observations or conclusions can be made:

1. The final clinical assessment of the 84 surgical cases shows 75% to be well or much improved, 22% to be improved, and 4% unchanged. Sixty-seven per cent are asymptomatic following resection.
2. Cases with clearly symptomatic, well-localized childhood bronchiectasis are ideal surgical candidates. Resection can be performed with almost no risks and a high expectancy for "cure."
3. Criteria for resection must be more selective in far advanced salvage cases, in those with bilateral disease, or where known residual disease is left after resection.
4. The isolated superior segment can be preserved in children with good results, provided there is clear bronchographic evidence that the *segment is entirely free of disease.*
5. When partially diseased segments, such as the superior segment of the lingula division of the left upper lobe, are retained and asked to fill a larger volume following extensive resection, there is a tendency for *even slightly altered bronchi to deteriorate postoperatively.*
6. Preservation of as much lung parenchyma as possible is important, even when this consists of only two or three segments to fill the hemithorax. The functional and cosmetic value of these retained segments has been demonstrated.
7. Bronchographic evidence of even full-blown, localized disease dose not justify resection unless the patient is clearly symptomatic.
8. In young children, bronchiectasis is often unstable. Resection can usually be safely postponed until an older age (usually 6 to 12 years).
9. In 111 patients not submitted to resection, symptoms have not changed (69%), or they have become worse (23%).
10. Review of this experience provides little data to justify the current bias against pulmonary resection for symptomatic, localized childhood bronchiectasis.

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