

Original Article

Surgical treatment of 60 patients with pulmonary malformations: What have we learned?*

Tratamento operatório de 60 pacientes com malformações pulmonares: O que aprendemos?

Altair da Silva Costa Júnior¹, João Aléssio Juliano Perfeito², Vicente Forte³

Abstract

Objective: To retrospectively analyze the medical charts of patients with pulmonary malformations submitted to surgical treatment and to investigate the clinical evolution prior to the definitive diagnosis. **Methods:** We analyzed the medical charts of patients with pulmonary malformations operated on at the São Paulo Hospital—Federal University of São Paulo/Paulista School of Medicine—from 1969 to 2004. Each medical chart was analyzed as to the following aspects: clinical profile; diagnosis; previous treatment; surgical treatment; and nosocomial complications. The inclusion criteria were having received a diagnosis of pulmonary malformation, having undergone pulmonary resection, and chart data being complete. **Results:** The analysis of the medical charts revealed that 60 patients diagnosed with pulmonary malformations—27 cases of bronchogenic cyst, 14 cases of congenital lobar emphysema, 10 cases of pulmonary sequestration, and 9 cases of cystic adenomatoid malformation—underwent surgery. Ages ranged from 4 days to 62 years (mean, 17.9 years). There was a predominance of males (55%). Ninety-two percent of the patients presented symptoms (mean duration, 15.37 months). Of the 60 patients undergoing surgery, 27 (45%) received preoperative home or hospital treatment with antibiotics. Regarding complications, we observed that morbidity was 23%, and mortality was 3.3%. Surgical times ranged from 1 to 8 h (mean, 3.2 h). **Conclusions:** Misdiagnosis or delayed diagnosis of pulmonary malformations resulted in unnecessary treatments and hospitalizations, as well as in frequent, recurrent infectious complications. We believe that the definitive treatment is surgery, which is curative and has low morbidity and mortality rates.

Keywords: Bronchogenic cyst; Bronchopulmonary sequestration; Cystic adenomatoid malformation of lung, congenital; Respiratory tract infections; Thoracic surgery.

Resumo

Objetivo: Analisar retrospectivamente os prontuários de pacientes com malformações pulmonares submetidos a tratamento operatório e verificar a evolução clínica até o diagnóstico definitivo. **Métodos:** Analisamos os prontuários dos pacientes com malformações pulmonares operados no Hospital São Paulo—Universidade Federal de São Paulo/Escola Paulista de Medicina—de 1969 a 2004. Cada prontuário foi analisado quanto aos seguintes aspectos: quadro clínico, diagnóstico, tratamento prévio, tratamento operatório e complicações hospitalares. Os critérios de inclusão foram os seguintes: ter diagnóstico de malformação pulmonar, ter sido submetido à ressecção pulmonar e ter prontuário com dados completos. **Resultados:** A análise dos prontuários revelou que 60 pacientes com diagnóstico de malformações pulmonares foram operados—27 casos de cisto broncogênico, 14 de enfisema lobar congênito, 10 de seqüestro pulmonar e 9 de malformação adenomatóide cística. A idade variou de 4 dias a 62 anos (média de 17,9 anos). Houve predominância do sexo masculino (55%). Noventa e dois por cento dos pacientes apresentavam sintomas (média de duração, 15,37 meses). Dos 60 pacientes operados, 27 (45%) receberam tratamento domiciliar ou hospitalar com antibiótico antes da operação. Quanto às complicações, observamos morbidade de 23% e mortalidade de 3,3%. A duração dos procedimentos operatórios realizados em nossos pacientes variou de 1 a 8 h (média, 3,2 h). **Conclusões:** A falha ou atraso no diagnóstico das malformações pulmonares resultou em tratamentos e hospitalizações desnecessárias e em complicações infecciosas recorrentes e freqüentes. Acreditamos que o tratamento definitivo é a operação, a qual é curativa e tem baixa morbidade e mortalidade.

Descritores: Cisto broncogênico; Seqüestro broncopulmonar; Malformação adenomatóide cística congênita do pulmão; Infecções respiratórias; Cirurgia torácica.

* Study carried out in the Thoracic Surgery Section of the Department of Surgery of the *Universidade Federal de São Paulo/Escola Paulista de Medicina* – UNIFESP/ EPM, Federal University of São Paulo/Paulista School of Medicine – São Paulo, Brazil.

1. Attending Physician in the Thoracic Surgery Section of the Department of Surgery. *Universidade Federal de São Paulo/Escola Paulista de Medicina* – UNIFESP/ EPM, Federal University of São Paulo/Paulista School of Medicine – São Paulo, Brazil.

2. Adjunct Professor in the Thoracic Surgery Section of the Department of Surgery. *Universidade Federal de São Paulo/Escola Paulista de Medicina* – UNIFESP/ EPM, Federal University of São Paulo/Paulista School of Medicine – São Paulo, Brazil.

3. Tenured Adjunct Professor in the Thoracic Surgery Section of the Department of Surgery. *Universidade Federal de São Paulo/Escola Paulista de Medicina* – UNIFESP/EPM, Federal University of São Paulo/Paulista School of Medicine – São Paulo, Brazil.

Correspondence to: Altair da Silva Costa Júnior. Disciplina de Cirurgia Torácica, Rua Napoleão de Barros, 715, 4º andar, Vila Clementino, CEP 04024-002, São Paulo, SP, Brasil.

Tel 11 5576-4295. Fax 11 5572-0448. E-mail: cirurgiatoracica@uol.com.br

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Introduction

Air-filled spaces in the lung were first described by Fontanus in 1638, whereas congenital cystic lung diseases were described in 1687 by Bartholinus and Marcellus Malpighius in the book "Opera Omnia". Congenital chest lesions are rare, and their clinical expression remains uncertain.⁽¹⁾

It is clear that the majority of alveolar morphogenesis occurs in the postnatal period. Morphometric studies estimate that only 8% (between 20 and 50 million) of the alveoli we possess in adulthood are present at birth. After the age of 8 years, the alveolar units continue to grow and develop, although more slowly. This growth and development is characterized by the increase in alveolar size (hypertrophy), known as compensatory lung growth. At the age of 9, there are approximately 280 million alveoli, a number close to that found in adulthood.^(2,3)

The incidence of lung malformations range from 30 to 42 cases for every 100,000 inhabitants per year—from 0.06 to 2.2% of the patients admitted to general hospitals.^(4,5) The forms of presentation of lung malformations vary. They can be very severe, including perinatal death, or remain totally asymptomatic until adulthood. It is important to recognize these conditions so that appropriate treatment (usually surgical) can be instituted, thereby avoiding unnecessary treatments.

The objective of the present study was to retrospectively analyze the medical charts of patients with pulmonary malformations submitted to surgical treatment and to investigate the clinical evolution prior to the definitive therapeutic option.

Methods

We retrospectively analyzed the medical charts of patients undergoing surgery for the correction of pulmonary malformations between 1969 and 2004 at the São Paulo Hospital—Federal University of São Paulo/Paulista School of Medicine—located in São Paulo, Brazil. The inclusion criteria were having received a diagnosis of pulmonary malformation (confirmed by anatomopathological examination), having undergone pulmonary resection, and having a chart in which the data were complete.

Each medical chart was analyzed as to the following aspects: clinical profile; diagnosis; previous treatment; surgical treatment; and nosocomial complications. The data evaluated were

divided into two categories: preoperative clinical data (age at surgery, gender, and infection prior to the definitive treatment, as well as presence and duration of symptoms); and postoperative data (extubation in the operating room, need for intensive care unit—ICU—care, surgical times, type of procedure performed, length of hospital stay, and complications).

This study was approved by the Ethics in Research Committee of the institution (CEP 0496/04).

Results

The analysis of the medical charts revealed that 60 patients diagnosed with pulmonary malformations—27 cases of bronchogenic cyst, 14 cases of congenital lobar emphysema, 10 cases of pulmonary sequestration, and 9 cases of cystic adenomatoid malformation—underwent surgery.

Preoperative clinical data

Ages ranged from 4 days to 62 years, with a mean of 17.9 years (Table 1). The incidence was highest in the first year of life (28.3% of the patients). Of the 60 patients, 32 (53.3%) underwent surgery at age 13 or younger (Figure 1).

There was a predominance of males: 33 (55%) males 27 (45%) females.

We observed that most (92%) of the patients presented symptoms that have little specificity, such as cough, dyspnea, or thoracic discomfort, but that were reason enough for these patients to seek medical attention (Table 2). The duration of symptoms was also evaluated. Despite the lack of specificity of the complaints, the duration reached up to 36 months in 3 patients, with a mean of 15.37 months (Table 1).

Table 1 – Age, duration of symptoms, surgical times, and length of hospital stay of the patients studied (n = 60).

Variable	Mean	Standard deviation
Age (years)	17.91	19.31
Duration of symptoms (months)	15.37	24.17
Surgical time (hours)	3.26	1.37
Length of hospital stay (days)	14.55	12.64

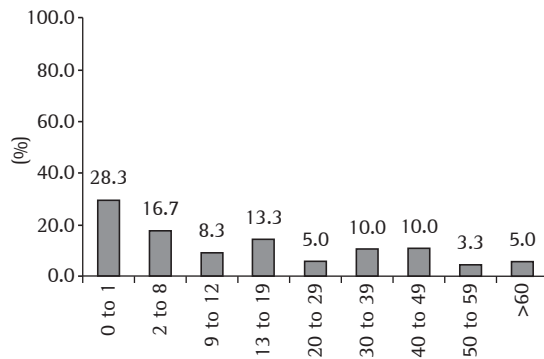


Figure 1 – Percentage distribution of the patients by age bracket (years).

A total of 27 (45%) of the patients received preoperative home or in-hospital treatment with antibiotics (Table 2). Recurrent pulmonary infection (pneumonia) was found in 18 (30%) of the patients. In 7 (11.7%) of the patients, more than five treatments were reported, and, in 11 (18.3%), two to four treatments were reported. One (1.7%) of the patients was treated with antibiotics on ten separate occasions, nine of which involved hospital admission, without ever being diagnosed with pulmonary malformation.

Postoperative data

Extubation at the end of the procedure, while the patient was still in the operating room, was performed in 58% of the cases. A total of 75% of the patients spent the immediate postoperative recovery

Table 2 – Incidence of symptoms, previous infection, extubation in the operating room, need for intensive care unit care, and complications (n = 60).

Variable	Yes	No
Symptoms	55 92%	5 8%
Previous infection	27 45%	33 55%
Extubation in the operating room	35 58%	25 42%
Need for ICU care	45 75%	15 25%
Complications	14 23%	46 77%

ICU: intensive care unit.

period in the ICU (Table 2). Surgical times ranged from 1 to 8 h, with a mean of 3.2 h (Table 1).

The procedures performed were as follows: pulmonary lobectomy (in 58%); cystectomy (in 13%); pulmonary segmentectomy (in 13%); sequestrectomy (in 10%); bilobectomy (in 3%); and pneumonectomy (in 3%). Length of hospital stay ranged from 3 to 70 days, with a mean of 14.55 days (Table 1).

Regarding complications, we observed that morbidity was 23% (14 patients, 4 of whom presented more than one complication). The types of complication were pneumonia (6 cases), atelectasis (6 cases), pleural empyema (3 cases), and sepsis (2 cases). Death occurred in 2 cases. One patient presented a retained clot and needed to undergo a second operation. The mortality rate was 3.3%—2 patients with malformations who underwent surgery before the age of 1 year, at different time points, and who developed atelectasis, pneumonia, and sepsis (one of them with hospital stay of 70 days).

Discussion

Pulmonary malformations have been known to physicians for more than three centuries. However, no consensus has yet been reached regarding how and when to treat these patients. Even if we consider that the incidence of pulmonary malformations is low, it is up to twice as high as the incidence of lung neoplasms—17.6 patients for every 100,000 inhabitants in the state of São Paulo.⁽⁶⁾

The alterations leading to malformations remain unknown. They are thought to occur between the second and sixteenth weeks of gestation, although there is still controversy in the literature.^(1,5,7)

When we think of congenital lung diseases, we relate them to asymptomatic patients. However, a review of the literature revealed that this does not reflect reality. In fact, malformations present symptoms in most patients.^(1,8-12) In a small percentage of patients, the disease will be identified in imaging studies performed routinely or based on another non-respiratory indication. Patients with pulmonary malformations will present symptoms at some point, typically before the age of 13. In the present study, 92% of the patients were symptomatic at the time of diagnosis. Our results are similar to those reported in other studies in the literature, in which

most such patients (65–100%) became symptomatic by the age of 15.^(8–12) Although the approach in asymptomatic patients remains a point of controversy, these lesions have been increasingly resected early due to the potential risk of complications.^(13,14) Even in asymptomatic patients, there is a risk of complications, and respiratory infection is the most common.

The prognosis of this disorder is unpredictable, and 45% of the patients with pulmonary malformations have been reported to have some type of complications prior to the definitive treatment. These complications would include hemoptysis, pneumothorax, esophageal compression, pleuritis, cyst infection, and pneumonia.^(15,16) Other complications that can occur are the development of malignant lesions (lung carcinoma or sarcoma), pneumothorax, hemoptysis, and hemothorax. Since pulmonary resection will eventually be necessary, it is best to perform the resection before such complications occur.^(13,17) One female patient in our sample underwent resection of an adenocarcinoma in the wall of the congenital cyst.

In our series, 27 (45%) of the patients needed home or hospital treatment due to previous pulmonary infections or to misdiagnosis. Those infections were characterized as pneumonia. One of those patients was hospitalized nine times prior to receiving a diagnosis of malformation. One group of authors showed that diagnosis was delayed for up to 7 years, with a mean of 1.6 years; the patients remained asymptomatic in this period, and all chest X-rays were abnormal.⁽¹⁸⁾ A review involving 320 children diagnosed with pneumonia emphasized the clinical-radiographic correlation with the evaluation of three professionals: a pediatric radiologist, a pediatric pulmonologist, and an emergency room pediatrician. The concordance among the observers was only 41%.⁽¹⁹⁾ In Brazil, the diagnosis of pneumonia, especially that known as “onset of pneumonia”, has evidently become commonplace. Many of the patients who present cough, rhinorrhea, and fever, as well as an abnormal X-ray, are (or have been) treated as having pneumonia. However, the X-ray finding could correspond to a malformation, such as bronchogenic cyst, and the clinical profile could correspond to that of an upper airway infection.

The most frequent misdiagnosis in patients with pulmonary malformations is pneumonia, and a

correct preoperative diagnosis is made in only 30% of the patients.⁽⁸⁾ Failure to diagnose malformations results in unnecessary treatments, recurrent infectious complications, and frequent hospitalizations. Even with a delayed diagnosis, all patients can be cured with surgical treatment. We believe that surgical treatment should not be performed in the acute phase of the respiratory infection, except in specific situations, with clinical treatment failure.⁽¹⁾

Surgical treatment is indicated due to risk of infection, interference with lung growth (ipsilateral or contralateral), heart failure, and progression of the pulmonary lesions to malignancy. During pregnancy, we can opt for clinical observation or prenatal interventions in fetuses with pleural effusion or hyperinflated cysts, which are usually treated with thoracocentesis. If necessary, intrauterine lobectomy can be used. In the postnatal period, the best treatment is malformation resection, which typically has an excellent prognosis.^(1,11,20,21)

In patients having experienced a great number of pulmonary infections prior to surgical treatment, the surgery itself is more difficult due to the inflammation around the malformation. There is a higher probability of finding pleural adhesences and fibrotic tissue around the lesion, and, therefore, surgical times can be longer. This probability is lower in asymptomatic patients. The reason for this difference in surgical times would be the longer duration of the disease, with more infections and inflammations in the lesion itself or in the adjacent parenchyma. With more inflammations and adhesences, resection tends to be slower and more cautious, and, therefore, surgical times are longer. In the literature, there have been reports of difficulties during surgery in patients who were symptomatic in the preoperative period.^(10,16,22) The degree of difficulty during surgery is higher in symptomatic patients than in asymptomatic ones, and surgical treatment should therefore be performed while patients are still asymptomatic.^(1,23)

Surgical treatment is recommended, and some authors recommend that lobectomy become the standard surgery.⁽²³⁾ Currently, lobectomy is preferred only in cases of malformations affecting the entire lobe. Whenever possible and safe, only the malformed area of the lung is resected, by anatomical or nonanatomical segmentectomy, as has been reported in the literature.^(24,25) However, lobectomy, as was used in the present study, is still the type

of resection most widely used for the treatment of pulmonary malformations.^(9,26-28)

In our sample, 58% of the patients were extubated at the end of the procedure in the operating room. A total of 75% required ICU care in the immediate postoperative period. The children required greater care in the postoperative period, which might have been a precaution rather than a real need. For patients with this type of congenital pulmonary lesion, usually young and without other comorbidities, if there are no complications during the procedure, postanesthesia recovery in the immediate postoperative period is sufficient, and there is no need for ICU admission.^(13,14,28)

Length of hospital stay has been a constant cause for concern, and the mean hospital stay in the present study was 14.55 days. It is important to emphasize that this retrospective study was long and subject to flaws. During the three decades of this study, medicine has been through many changes. Currently, diagnostic investigation is preferably performed in outpatient clinics, reducing the length of hospital stays.

A total of 23% of the patients presented complications, and mortality was 3.3%. Among the complications, atelectasis prevailed (in 5 children and 1 adult), especially in the patients younger than 2 years, which is explained by the fact that the airway diameter is too small for the secretions, which are sometimes as viscous as those in adults. When the patients were divided by age bracket, morbidity and mortality were found to be higher in the children younger than 1 year (58% of such patients had atelectasis and pneumonia; 11.8% died). It is worthy of note that, in patients in this age bracket, indications for surgery should be pondered whenever permitted by the clinical profile. If surgical treatment can be postponed until the child is 1 year of age or older, morbidity and mortality rates will be lower. We have found similar incidences in the literature, with morbidity being 40% and mortality being around 10%. Among the most common complications reported are prolonged drainage, empyema, pneumonia, and infection at the incision site.^(11,16,29) In the present study, the morbidity and mortality rates were much lower than those reported in the literature. It should be borne in mind that the complications that can occur prior to surgical treatment, caused by the malformation itself, reach

up to 45% of the patients,⁽¹⁶⁾ an incidence that is higher than the postoperative morbidity.

There is no consensus in the world literature regarding the ideal age at which surgical treatment should be performed after birth. There are many reasons why such surgery should be performed in adults rather than in children, and vice versa. However, it is known that lung growth through hyperplasia (actual increase in the number of alveoli) occurs up to the age of 8 years. Up to the age of 9 years, there is lung growth in volume and size, but not in the absolute number of alveoli.⁽¹⁾ In a study published in 1987, 230 patients submitted to pneumonectomy were monitored for up to 30 years. At the time of surgery, the patients were divided by age bracket. In the group undergoing surgery between birth and 5 years of age, 96% maintained total lung capacity even after pneumonectomy.⁽³⁰⁾ Other authors recommend that surgical treatment of patients diagnosed with pulmonary malformations be performed at between 3 and 6 months of age so that lung growth can occur.^(13,29,30)

We conclude that pulmonary malformations were underdiagnosed at our facility. Sometimes, several conditions were confused with pulmonary infections, and, sometimes, they were treated as pneumonia. The diagnostic hypothesis of congenital pulmonary abnormality should be, at least, considered by the physician, especially in patients with recurrent pneumonia. We reiterate that misdiagnosis or delayed diagnosis of pulmonary malformations resulted in unnecessary treatments and hospitalizations, as well as in frequent, recurrent infectious complications. We believe that the definitive treatment is surgery. There is a tendency toward resection being as conservative and as safe as possible. Currently, the indication for lobectomy is limited to patients in whom the entire lobe is affected. In patients who are still asymptomatic but present imaging evidence of pulmonary malformation, surgery should be indicated even without confirmation of the diagnosis. If possible, we prefer to perform surgical treatment when the patient is ≥ 1 year of age. We emphasize that the surgical treatment is curative, with low rates of morbidity and mortality.

Further studies are necessary in order to elucidate controversies and principally to enlighten the general medical population about the importance of the correct diagnosis of these diseases.

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