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Treatment of pulmonary sequestration in children: surgical resection versus endovascular embolization

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Abstract

Introduction: Pulmonary sequestration is a lower respiratory malformation of the tract, which consists of a nonfunctional mass of lung tissue without communication with the tracheobronchial tree and the blood supply from one or more aberrant systemic arteries. This study aimed to analyze the outcomes of pediatric patients with pulmonary sequestration treated with surgical resection (thoracotomy, video-assisted thoracoscopy) or endovascular embolization.

Methodology: a descriptive, observational, retrospective, cross-sectional study in a tertiary pediatric hospital in Ecuador from January 2017 to December 2022. Data were collected from chart review.

Results: 9 patients under the age of 18 were included, with a diagnosis of pulmonary sequestration, six females and three males, five were treated with surgery, 4 (44%) by thoracotomy and 1 (11%) by video-assisted thoracoscopy, 4 (44%) with endovascular embolization, the mean age of patients who underwent surgery was 41.36 months (range: 0.1-144 months), and of those who underwent embolization was 12.9 months (range: 0.6-41 months). One patient died six days after the surgical resection of the pulmonary sequestration by thoracotomy secondary to pulmonary hypertension due to his underlying heart disease. One patient treated with endovascular embolization developed a decrease in pedal pulse, which resolved 24 hours after starting streptokinase.

Conclusion: In this study, surgery and endovascular embolization were safe procedures; however, no significant differences were found between both treatment options; more prospective studies are needed that include a large population sample of patients treated by endovascular embolization.

Keywords:

MESH: Child; Pulmonary sequestration; Thoracotomy; Videothoracoscopy; Endovascular embolization.

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Introduction

Pulmonary sequestration (PS) is a malformation of the lower respiratory tract, consisting of a nonfunctional mass of lung tissue without communication with the tracheobronchial tree, and its blood supply comes from one or more aberrant systemic arteries [1]. Two types of PS have been described, intralobar pulmonary sequestration (IPS) and extralobar pulmonary sequestration (EPS); in both cases, an aberrant systemic artery provides the blood supply, and the IPS is found within the normal lung parenchyma sharing the same visceral pleura. Its blood drainage is into the pulmonary veins, and the EPS has its visceral pleura. Its drainage is into the systemic veins [2].

SP represents approximately 0.15% to 6.40% of all congenital pulmonary malformations [3]. Several locations have been described, but it is well known that more than 90% are located in the chest, and less than 10% are located below the diaphragm [4].

Several therapeutic options have been described over the years for PS, such as surgical resection of the sequestered lobe or segment by thoracotomy or video-assisted thoracoscopy, and endovascular embolization (EE) has also been described as an alternative treatment to surgery [5, 6].

There is currently no consensus on the best therapeutic option for PS in children, and the management of asymptomatic patients with PS as an incidental finding is still being discussed [7].

This study aims to analyze the results of pediatric patients with PS diagnosed in our center who were treated with surgical resection (extended thoracotomy, video-assisted thoracoscopic) or endovascular embolization.

Population and methods

Design of the investigation

This is a retrospective, cross-sectional, observational study.

Scenery

The study was carried out in the pediatric surgery service of the Dr. Roberto Gilbert Elizalde Children's Hospital of the Guayaquil Benevolent Board in Guayaquil-Ecuador. The study period was from January 1, 2017, to January 31, 2022.

Inclusion criteria

Hospitalized pediatric patients with an established diagnosis of congenital bronchopulmonary sequestration entered the study. The tenth edition (ICD-10) international classification of diseases was used for the research: Q33.2. Incomplete records were excluded from the analysis.

Studio size

The universe was patients admitted to the institution. The sampling was nonprobabilistic for convenience, and all possible cases were included in the study period.

Variables

The variables were dependent on the type of surgical intervention: group 1: surgery, group 2: endoscopic surgery. Independent variables: sex, age, weight, symptoms, associated anomalies, characteristics of pulmonary sequestration, operative time, length of hospital stay, complications, and mean follow-up time.

Data sources/measurement

The data were collected in a specific electronic form for this purpose. The hospital's electronic system of clinical records was used to investigate cases. The results of successful treatment of patients treated through ES were evaluated with control CT and classified as follows: Good (B) when the absence of the lesion was evident; Intermediate (I) when there was evidence of a reduction in the largest diameter of the lesion more than 30%; Bad (M) when there was evidence of a reduction in the largest diameter of the lesion of less than 30% or an increase in size as recommended in the study by Cho et al. [8].

To perform the EPS, the femoral artery was used, and aortography was performed to identify the aberrant vessel, where it was shown that in all cases, they were dependent on the abdominal aorta and the venous drainage of the PS. Then, the 4 Fr catheter was selectively catheterized. An aberrant vessel was occluded with an Amplatzer™ occluder device.

In cases of open surgical resolution, a posterolateral thoracotomy was performed on the affected side with resection of the lesion.

In cases of surgery by video-assisted thoracoscopy, the patient was placed in lateral decubitus.

Three 5-mm ports were placed, and dissection and division of the vessels were performed with LigaSure® laparoscopic forceps. Then, the sample was placed in the collection bag, morcellation of the sample was performed, and it was subsequently extracted through one of the ports.

In all surgeries, both open and video-assisted, a chest tube connected to a water seal was placed, and once the chest X-ray showed good lung expansion, the chest tube was removed.

Statistical method

The data analysis is univariate and descriptive with frequencies and percentages. A second bivariate analysis compares the results of the group of patients treated by a plastic surgeon vs. a pediatric surgeon. A comparison of proportions with Chi^2 is used. The statistical package SPSS v.21 (version 2012, Armonk, NY: IBM Corp.) was used for the analysis.

Results

A total of 228 cases were analyzed (Figure 1).

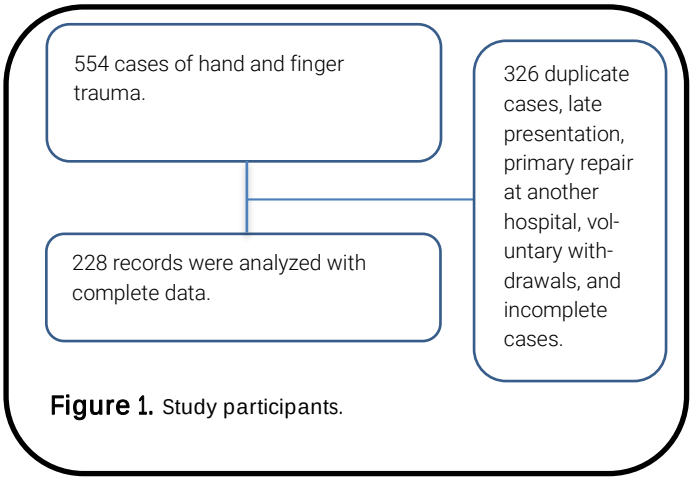

Figure 1. Study participants.

General characteristics of the study group

Four patients were diagnosed with SP through computerized axial tomography, in which EE was performed; five patients were diagnosed based on the pathological results after surgery; four patients underwent SP resection due to thoracotomy; and one patient underwent video-assisted thoracoscopy. The results of endovascular procedures and surgical resections were compared.

Nine patients were included, six female and three male; five patients were treated with surgery, 4 (44%) by thoracotomy, 1 (11%) by video-assisted thoracoscopy, and 4 (44%) patients were treated with EPS. Table 1 shows a summary of the characteristics and their clinical evolution of all the patients by group.

The average age at the time of surgery was 41.36 months (range: 0.1-144 months), and in the case of patients treated with EPS, it was 12.9 months (range: 0.6-41 months). At the time of surgery, the average weight of the patients was 12.04 kg (range: 1.7-36.6 kg), and for those treated through EPS, it was 6.93 kg (range: 3.4-13 kg). The symptoms presented by the patients before treatment were respiratory distress in 5 patients, four asymptomatic.

The associated anomalies found in the patients treated with surgery were one (20%) patient with dextrocardia and patent ductus arteriosus and one (20%) patient with patent small ductus arteriosus. In the case of the group treated by EE, it was one with tetralogy de Fallot, hypoplasia of the left ventricle (25%), and one (25%) patient with partial anomalous drainage of the pulmonary veins; in all the patients, once the PS was treated, the associated pathologies were resolved.

In the group of patients treated by surgery, it was observed that 4 (80%) patients presented left PS and 1 (20%) right, 3 (60%) were extralobar and 2 (40%) intralobar, in the group treated by EE 4 (80%) were extralobar and 1 (20%) intralobar, two were right, one left, and one patient presented right and left infra-diaphragmatic PS. All thoracic PSs were found at baseline.

The average operative time used in the group of patients treated by thoracotomy was 127.5 minutes (range: 70-295 minutes), and the only case performed by video-assisted thoracoscopy was 120 minutes; in the case of the group treated with EE, the average was 190 minutes (range: 125-300 minutes).

The average time of use of the chest tube after surgery was 4.6 days (range 4-6 days), and none of the patients treated with EE required a chest tube. In the case of the group treated with surgery, the average hospital stay after the procedure was 10.75 days (range: 6-21 days), and for the group treated with EE, it was nine days (range: 1-22 days). Only 1 (25%) patient died six days after PS resection by thoracotomy due to severe pulmonary hypertension due to underlying

heart disease, and 1 (25%) patient had a decreased pedal pulse after EE, which improved 24 hours after starting streptokinase.

Patients were followed up for an average of 30 months (range: 6-48 months) for those treated by surgery and 42 months (range: 36-60 months) for those treated by the US.

Table 1. Demographic characteristics of the patients.

	Surgery (n=5)	EE (n=4)	*P*
Female Male	3:2	3:1	1
Age (months)	41.4 (0.1-144)	12. (0.6-41).	1
Weight (kg)	12.04 (1.7-36.6)	6.93 (3.4-13).	0.806
Symptoms			
Symptomatic	3	2	-
asymptomatic	2	2	-
Associated anomalies			
Yes	2	2	-
No	3	2	-
PS side			
Left	4	2	-
Law	1	3	-
PS Type			
Extralobar	3	4*	-
Intralobar	2	1	-
Operative times			
Operative time (minutes)	127.5 (70-295 min)	190 (125-300)	0.085
Length of hospital stay (days)	10.8 (6-21)	9.0 (1-22)	0.806
Follow-up time (months)	30 (6-48)	42 (36-60)	0.283
Complications			
Complications	1	1	-

* one patient presented two infra diaphragmatic pulmonary sequestrations; ** one patient was treated by video-assisted thoracoscopy. EE: endovascular embolization.

Treatment type

The type of repair was classified as follows: 1. Nonsurgical management: Nine patients (3%) required analgesia, two patients (1%) underwent drainage of subungual hematomas, and 22 patients (9%) were managed with dressings of minor injuries. 2. Surgical management included simple sutures for soft tissue injuries in 85 patients (37%), nail repositioning under general anesthesia in 3 patients (1%), and complex sutures with nail repair in 62 patients (27%). Local flaps were performed in 42 patients (18%), the most common being the VY flap preferred by plastic surgeons (83%). Three patients who presented total amputations underwent stump fabrication (1%) (Table 1).

Associated distal phalanx fractures were stabilized using percutaneous fixation, mostly with hypodermic needles or immobilization of the affected finger. Eighty-four patients (36%) required outpatient follow-up appointments within 7 to 30 days after discharge; long-term results were not available in this study. Eight patients (3%) presented complications associated with extensive areas of injury, and tissue compromise, such as flap necrosis, was re-explored (Table 1).

Discussion

The prevalence of congenital pulmonary malformations is 1 in 8,000 to 35,000 live births, and SP is a variant of this, with an estimated incidence of 0.15 to 6.4% [3, 9]. Many hypotheses have been described on the etiology of PS; however, the most accepted is where it is described as an accessory pulmonary bud that forms between 4 and 8 weeks of gestation and results in pulmonary malformation [10].

Several complications of PS have been described, the most common being infection, recurrent pneumonia, respiratory distress, fetal chylothorax, heart failure, pleural effusion, hemoptysis, pulmonary hypertension, and the development of malignancy. Associated malformations such as malformations have also been reported. The most common association is communicating bronchopulmonary foregut, bronchogenic cysts, cystic adenomatoid malformations, scimitar syndrome, and diaphragmatic hernia [7, 8].

In other published series, the percentage of asymptomatic patients with PS ranged from 10 to 15% [11]. However, in our series, the percentage of asymptomatic patients was 40% for the group that was treated by surgery and 50% for those who were treated for EE; in both groups, despite being asymptomatic, the decision was made to be treated to avoid future complications as a result of PS.

The average time to perform PS resection by thoracotomy published in the series by Cho et al. [8] was 132.9 minutes. In the case of the thoracoscopic route, it was 151.9 minutes, similar to our series with an average of 126 minutes for thoracotomies; only one thoracoscopic resection was performed, which lasted

120 minutes; in the case of the group treated by EE, the average of the procedure was 190 minutes; however, it is worth noting that one patient presented 2 PS.

In our series, the average hospital stay after surgery was 10.75 days, similar to that published in various series where the average hospital stay after surgery ranged from 7.9 to 8.1 days [5, 12]. In the case of the group treated with EE in our series, the average stay was nine days, which differs from other published series [6, 13] in which the average ranged between 2.3 and 2.6 days; however, 50% of the patients treated in our EE series presented associated complex heart disease.

According to the literature reviewed, the treatment of PS by surgery and by EE is safe in children with a low percentage of complications [6, 8, 14], which was observed in our series since only one patient died six days after surgery due to pulmonary hypertension, and one patient treated for EE presented a decreased pedal pulse, which improved 24 hours after starting streptokinase.

Forty percent of asymptomatic patients were treated through a thoracotomy, and 50% of asymptomatic patients were treated through EE. Between 10-33% [7, 8], a consensus has not yet been described on whether asymptomatic patients should be operated on, but according to our results, treated asymptomatic patients presented a good evolution with both therapeutic options.

The average follow-up for patients undergoing surgery was 30 months with good evolution and 42 months for those treated for EE, in whom a good evolution was evidenced since complete regression of the PS was obtained, confirmed by a control chest tomography. In other series [7, 8], the mean follow-up ranged from 8.3 to 71.2 months.

The main limitation of this study is its retrospective nature, and the number of patients included is limited. Therefore, prospective studies with a larger number of patients are recommended to establish which is the best therapeutic option.

Conclusions

In this study, surgery and EE were safe procedures; however, no significant differences were found between both therapeutic options, and studies with a larger population and prospective characteristics are necessary.

Abbreviations

CT: computerized axial tomography.
PS: Pulmonary sequestration.
EPS: extralobar pulmonary sequestration.
ILS: intralobar lobar sequestration.
EE: endovascular embolization.

Supplementary information

None declared by the authors.

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Author contributions

Luis Guillermo Mendoza Saldarreaga: Conceptualization, Data Retention, Funding, Research, Resources, Software, Writing - original draft.
Ernesto Santiago Fabre Parrales: Conceptualization, data conservation, supervision, acquisition of funds, research, resources, and writing: revision and editing.
Jorge Alejandro Oliveros-Rivero: Data curation, research, acquisition of funds, Supervision, Methodology.
Julian Luna Montalvan: Conceptualization, Data Conservation, Supervision, Visualization, Methodology.
Daniel Acosta-Farina: Conceptualization, data retention, fundraising, research.
All authors read and approved the final version of the manuscript.

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Availability of data and materials

The data sets generated and analyzed during the current study are not publicly available due to participant confidentiality but are available through the corresponding author upon reasonable academic request.

Statements

Ethics committee approval and consent to participate

The Ethics Committee approved the research protocol of the Teaching Committee of the Dr. Roberto Gilbert Elizalde Children's Hospital.

Publication consent

The authors have permission for publication by the guardians of patients who appear in the photographs duly unidentified.

Conflicts of interest

The authors declare no conflicts of interest.

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