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RESEARCH ARTICLE

Congenital pulmonary malformations in children in a pediatric hospital in Peru, 2010-2020

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Abstract

Background: *Congenital pulmonary malformations (CPMs) are rare in children. This study aimed to describe the clinical, imaging characteristics, and treatment of patients with this pathology.* **Methods:** *We conducted a descriptive and retrospective study with data from patients with CPMs diagnosed at Instituto Nacional de Salud del Niño-Breña (Lima-Peru), from January 2010 to December 2020. We described CPM clinical and imaging characteristics, type and treatment.* **Results:** *The sample was formed of 70 patients. The median age was 29 months (range 15 days-14 years) and the male/female ratio was 1.4. The chest tomography found parenchymal involvement in 50 (71.4%) cases and mixed involvement (parenchymal and vascular) in 18 (25.7%) cases. Congenital malformation of the pulmonary airway was present in 39 (55.7%) cases, followed by bronchogenic cyst in 10 (14.3%), intralobar pulmonary sequestration in 9 (12.9%), and extralobar pulmonary sequestration in 7 (10%). Lobectomy was performed in 61 (87.1%) cases, cystectomy in 5 (7.1%), segmentectomy in 2 (2.9%), and embolization in 2 (2.9%). The most frequent post-operative complication was pneumonia, found in 9 (12.9%) cases. The median hospital stay was 26 days. No patient died during hospitalization.* **Conclusions:** *In our institution, the most frequent CPM was congenital malformation of the pulmonary airway, and lobectomy was the most frequently performed surgical procedure. CPMs represent a diverse group of disorders of lung development with varied imaging patterns and clinical manifestations.*

Keywords: *Congenital pulmonary malformations. Congenital malformation of the pulmonary airway. Pulmonary sequestration. Bronchogenic cyst. Congenital lobar emphysema. Children. Peru.*

Malformaciones pulmonares congénitas en niños de un hospital pediátrico de Perú, 2010-2020

Resumen

Introducción: *Las malformaciones pulmonares congénitas son poco frecuentes en niños. El objetivo de este estudio fue describir las características clínicas, imagenológicas y tratamiento de los pacientes con esta patología.* **Métodos:** *Se llevó a cabo un estudio descriptivo y retrospectivo con datos de los pacientes con malformaciones pulmonares congénitas diagnosticados en el Instituto Nacional de Salud del Niño-Breña (Lima-Perú) entre enero 2010 y diciembre 2020. Se describieron las características clínicas, imagenológicas, el tipo de malformación pulmonar congénita y el tratamiento.* **Resultados:** *La muestra fue de 70 pacientes. La mediana de edad fue 29 meses, la relación masculino/femenino fue 1.4. En la tomografía*

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Date of reception: 8-04-2023
Date of acceptance: 13-07-2023
DOI: 10.24875/BMHIM.23000055

Available online: 01-09-2023
Bol Med Hosp Infant Mex. 2023;80(4):235-241
www.bmhim.com

de tórax se encontró compromiso parenquimal en 50 (71.4%) casos, y compromiso mixto (parenquimal y vascular) en 18 (25.7%). La malformación congénita de la vía aérea pulmonar se observó en 39 (55.7%) casos, seguida del quiste broncogénico en 10 (14.3%), secuestro pulmonar intralobar en 9 (12.9%) y secuestro pulmonar extralobar en 7 (10%). La lobectomía fue realizada en 61 (87.1%) casos, la quistectomía en 5 (7.1%), segmentectomía en 2 (2.9%) y embolización en 2 (2.9%). La complicación posquirúrgica más frecuente fue la neumonía en 9 (12.9%) casos. La mediana de estancia hospitalaria fue de 26 días. Ningún paciente falleció durante la hospitalización. **Conclusiones:** *En nuestra institución, la malformación pulmonar congénita más frecuente fue la malformación congénita de la vía aérea pulmonar, y la lobectomía el procedimiento quirúrgico más comúnmente realizado. Las malformaciones pulmonares congénitas representan un grupo diverso de trastornos del desarrollo pulmonar con variados patrones imagenológicos y manifestaciones clínicas.*

Palabras clave: *Malformaciones pulmonares congénitas. Malformación congénita de la vía aérea pulmonar. Secuestro pulmonar. Quiste broncogénico. Enfisema lobar congénito. Niños. Perú.*

Introduction

Congenital pulmonary malformations (CPMs) are heterogeneous disorders that occur during lung development and affect pulmonary airways, parenchyma, or pulmonary vasculature. The most common CPMs are congenital pulmonary airway malformation (CPAM), pulmonary sequestration, congenital lobar emphysema, and bronchogenic cysts[1-3].

The estimated incidence of CPMs is 30-42 cases/100,000 population/year, with approximately 10% of cases diagnosed at birth and 14% diagnosed during childhood and adolescence[1,4-6].

The clinical presentation varies from severe conditions, such as recurrent bronchopulmonary infections in children, to completely asymptomatic clinical situations in adolescents and adults. The latter are diagnosed incidentally when chest imaging is performed as part of other studies[1,5,7].

Diagnostic support techniques during the prenatal period include maternal-fetal ultrasonography. Prenatal magnetic resonance imaging (MRI) is required if the lesions are extensive enough to cause pulmonary hypoplasia. In the postnatal period, regardless of the clinical presentation, a delayed diagnosis requires chest radiography, computed tomography (CT), or MRI, which are useful to confirm and characterize the different CPMs to follow a correct surgical approach[7-12].

A few studies on CPMs have been conducted in Latin America[5,13,14], but in Peru, no experience in diagnosing and managing this pathology in children has been reported. In the Instituto Nacional de Salud del Niño-Breña (INSN-Breña), the reference center for pediatric pathologies in Peru, diseases of the respiratory system ranked third among the 20 leading causes of hospitalization during the period 2010-2020. In general, congenital malformations were reported in 9.2% of patients, although the frequency of CPMs was not reported[15].

Since information on this pathology in children is still limited, this study aimed to describe the clinical, imaging characteristics, and management of patients with CPMs in a pediatric hospital in Peru between 2010 and 2020.

Methods

Design and population

We conducted a descriptive and retrospective study. Data were collected from the medical records of patients diagnosed with CPM at the INSN-Breña in Lima, Peru, between January 2010 and December 2020. The INSN-Breña is an institution of the Peruvian Ministry of Health (MINSA, for its Spanish acronym) that carries out health care and research activities[16] and is a national reference center for childhood diseases.

The study population included 70 patients. The diagnosis of CPM was a combination of clinical and imaging findings and anatomopathological confirmation. Patients with CPM diagnosis and who underwent surgery with confirmatory anatomopathological analysis were included in the study. We excluded ten deceased patients with suspected CPM but no anatomopathological confirmation.

Study variables

Clinical variables such as age at diagnosis (months), sex, origin, clinical manifestations, and saturation on admission were described. The imaging variables were chest radiography (focal hyperlucency, focal opacity, and multicystic mass) and tomographic features (parenchymal, mediastinal, and mixed involvement). The type of CPAM was reported according to the classification proposed by Stocker[17]. The type of CPMs, surgical

procedure, post-operative complications, and hospital stay were also described.

A data collection form was used to obtain information from medical records, the hospital imaging database (PACS digitized system), and the anatomic pathology report.

Statistical analysis

For descriptive analysis, numerical variables were summarized as median and range Categorical variables were expressed as absolute and relative frequencies.

The difference between males and females was evaluated for bivariate analysis. The non-parametric Mann–Whitney U-test was used for continuous variables, and Fisher's exact test for categorical variables, with $p < 0.05$ considered statistically significant. All analyses were performed with the Statistical Package for the Social Sciences® for Windows version 24 (IBM, Chicago, IL).

Ethical aspects

The research was approved by the Ethics and Research Committee of the INSN-Breña (Nº 036-2022-CIEI-INSN). The present study was based on Helsinki's ethical principles. Data confidentiality was respected using a numerical code to avoid identifying the participants.

Results

During the study period, 70 cases of CPMs were reported. The median age was 29 months (range 15 days to 14 years), and the male sex was more common with 41 (58.6%) cases. Forty-four (62.9%) patients were from Lima, and the rest were from other departments of Peru (Figure 1).

The most common clinical manifestations were recurrent infections in 28 (40%) cases and respiratory distress in the neonatal period in 19 (27.1%) cases. Twenty-two patients (31.4%) were asymptomatic at diagnosis. On chest radiography, focal hyperlucency was the most common feature (32 [45.7%] cases), and on tomography, parenchymal involvement (50 [71.4%] cases) was observed.

The most common type of CPM was CPAM, with 39 (55.7%) cases, followed by pulmonary sequestration, with 16 (22.9%) cases. Three (4.3%) cases were mixed CPM (case 1: extralobar pulmonary sequestration and type 2 CPAM in the right lower lobe; case 2: intralobar pulmonary sequestration and type 2 CPAM in the left lower lobe; and case 3: intralobar pulmonary sequestration and type 2 CPAM in the left upper lobe) (Table 1). The most common site of CPM was the left lower lobe, with 21 (30%) cases, followed by the right lower lobe, with 20 (28.6%) cases.

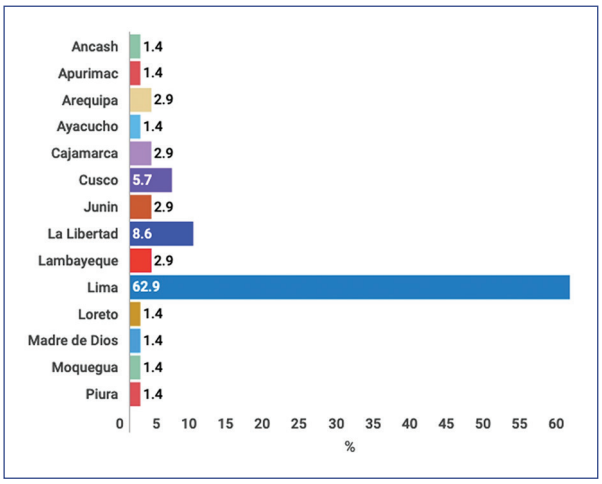

Figure 1. Origin of patients with congenital pulmonary malformations.

Lobectomy was the most common surgical procedure, with 67 (87.1%) cases; pneumonia was the most common post-operative complication, with 9 (12.9%) cases. The median hospital stay was 26 days (range 2-135 days) (Table 2).

No differences were observed between males and females concerning the variables evaluated ($p > 0.05$), except for chest CT findings ($p = 0.018$) (Tables 1 and 2). According to Stoker's classification, type 1 CPAMs were the most common, followed by type 2 CPAMs (Table 3).

No patient died during hospitalization, and all were discharged clinically stable.

Discussion

Several theories have attempted to explain the origin of CPMs: defective sprouting of the tracheobronchial tree at 24-36 weeks of gestation, obstruction of the developing bronchus, pulmonary vascular anomalies, and genetic alteration of signaling pathways for lung formation[1,7,18].

The most common CPMs are CPAM, bronchopulmonary sequestration, congenital lobar emphysema, and bronchogenic cysts[1,3,19,20]. After reviewing a series of cases of 11-year-old patients who underwent surgery

Table 1. Clinical and radiological characteristics and location of congenital pulmonary malformations by sex

Characteristics	Total n = 70 (%)	Males n = 41 (%)	Females n = 29 (%)	p-value
Age (months)+	29 (0.5-168)	20 (0.5-168)	29 (0.8-168)	*0.99
Clinical manifestations				**0.213
Asymptomatic	22 (31.4)	13 (31.7)	9 (31.0)	
Neonatal respiratory distress	19 (27.1)	14 (34.1)	5 (17.2)	
Recurrent infections	28 (40.0)	13 (31.7)	15 (51.7)	
Hemoptysis	1 (1.4)	1 (2.4)	0 (0.0)	
$SatO_2$ to FiO_2 0.21	97 (92-99)	97 (92-99)	97 (94-99)	*0.912
Chest X-ray				**0.340
Focal hyperlucency	32 (45.7)	18 (43.9)	14 (48.3)	
Focal opacity	28 (40.0)	19 (46.3)	9 (31.0)	
Multicystic mass	10 (14.3)	4 (9.8)	6 (20.6)	
Chest CT				**0.018
Parenchymal involvement	50 (71.4)	26 (63.4)	24 (82.8)	
Mediastinal involvement	2 (2.9)	0 (0.0)	2 (6.9)	
Mixed involvement (parenchymal + vascular)	18 (25.7)	15 (36.5)	3 (10.3)	
Type of CPM				**0.306
CPAM1	23 (32.9)	12 (29.3)	11 (37.9)	
CPAM2	12 (17.1)	7 (17.1)	5 (17.2)	
CPAM3	2 (2.9)	2 (4.9)	0 (0.0)	
CPAM4	2 (2.9)	0 (0.0)	2 (6.9)	
Bronchogenic cyst	10 (14.3)	4 (9.8)	6 (20.7)	
CLE	2 (2.9)	1 (2.4)	1 (3.4)	
Extralobar PS	7 (10.0)	6 (14.6)	1 (3.4)	
Intralobar PS	9 (12.9)	7 (17.1)	2 (6.9)	
Mixed CPM	3 (4.3)	2 (4.9)	1 (3.4)	
Localization				**0.182
RUL	15 (21.4)	11 (26.8)	4 (13.8)	
ML	4 (5.7)	2 (4.9)	2 (6.9)	
RLL	20 (28.6)	9 (22.0)	11 (37.9)	
LUL	5 (7.1)	3 (7.3)	2 (6.9)	
LLL	21 (30.0)	15 (36.6)	6 (20.7)	
Md	5 (7.1)	1 (2.4)	4 (13.8)	

*Mann–Whitney U-test.
**Fisher's exact test.
+median (range).
CLE: congenital lobar emphysema; CPAM - congenital pulmonary airway malformation; CPM: congenital pulmonary malformation; CT: computed tomography; FiO_2: inspired fraction of oxygen; LLL - left lower lobe; LUL, left upper lobe; Md: mediastinum; ML, middle lobe; PS: pulmonary sequestration; RLL, right lower lobe; RUL, right upper lobe; $SatO_2$: oxygen saturation.

in our institution, 70 CPMs were identified: 39 (55.7%) cases corresponded to CPAM, 16 (22.9%) to pulmonary sequestration, 10 (14.3%) to bronchogenic cyst, 3 (4.3%) to mixed congenital malformations, and 2 (2.9%) to congenital lobar emphysema.

CPAMs account for about 30-40% of all CPMs. Stocker classified them into five types, with the following proportions: type 0 CPAM < 2%, type 1 CPAM: 60-70%, type 2 CPAM: 15-20%, type 3 CPAM: 5-10%, and type 4 CPAM < 10%[17]. The proportions found in our study were similar to those reported by Stocker (Table 3).

The clinical manifestations of CPMs are varied; some cause perinatal respiratory failure, which can be very severe, others cause recurrent bronchopulmonary infections in the child, and others may be asymptomatic until adolescence and diagnosed incidentally in the course of other studies[1,4,5]. In this report, the most common clinical manifestations were recurrent bronchopulmonary infections and neonatal respiratory distress; however, approximately one-third of the patients were asymptomatic.

CPMs are diagnosed based on clinical and imaging findings, and confirmed with pathologic evidence. Frontal and lateral chest radiographs show focal hyperlucency, focal opacities, focal cystic masses, vascular abnormalities, airway abnormalities, or thoracic asymmetry[7,8,18]. We observed these features in our cases, with focal hyperlucency being the most frequent finding.

Table 2. Surgical management, post-surgical complications, and hospital stay of patients with congenital pulmonary malformations

Characteristics	Total n = 70 (%)	Males n = 41 (%)	Females n = 29 (%)	p-value
Surgical procedure				**0.733
Lobectomy	61 (87.1)	37 (90.2)	24 (82.8)	
Segmentectomy	2 (2.9)	1 (2.4)	1 (3.4)	
Cystectomy	5 (7.1)	2 (4.9)	3 (10.3)	
Embolization	2 (2.9)	1 (2.4)	1 (3.4)	
Complications				**0.961
Pneumonia	9 (12.9)	5 (12.2)	4 (13.8)	
Pulmonary abscess	1 (1.4)	0 (0)	1 (3.4)	
Pulmonary air leak	1 (1.4)	1 (2.4)	0 (0)	
Atelectasis	4 (5.7)	3 (7.3)	1 (3.4)	
Pneumothorax	5 (7.1)	3 (7.3)	2 (6.9)	
Residual abscess	2 (2.9)	1 (2.4)	1 (3.4)	
None	48 (68.6)	28 (68.3)	20 (69.0)	
Hospital stay (days)+	26 (2-135)	24 (4-135)	28 (2-72)	*0.747

*Mann–Whitney U-test.
**Fisher's exact test.
+median (range).

Table 3. Frequency of congenital pulmonary malformations studied according to Stocker classification[17]

CPAM type	Histological features	n = 39 (%)
0	Involvement of all lung lobes, stillborn	0 (0.0)
1	Single or multiple cysts > 2 cm, pseudostratified columnar epithelium	23 (59.0)
2	Single or multiple cysts < 2 cm, columnar or cuboidal epithelium	12 (30.8)
3	Predominant solid lesions, < 0.5 cm, cuboidal epithelium	2 (5.1)
4	Large air-filled cysts, flattened epithelial cells	2 (5.1)

CPAM: congenital pulmonary airway malformation.

High-resolution tomography with multiple 2D and 3D reconstructions provides good-quality images of the airways, lung parenchyma, and vasculature, and the use of contrasting material provides a better approximation of CPM. Because children are radiosensitive, one option is to use MRI, although its ability to evaluate lung parenchymal abnormalities is limited[7,10,11,20,21]. In this study, the 70 cases underwent chest CT and parenchymal involvement was found in 50 (71.4%) and mixed involvement (parenchymal and vascular) in 18 (25.7%). Due to the lack of equipment for this study in our institution, MRI was not performed.

Other supportive diagnostic tests of CPMs during pregnancy include Doppler ultrasound and MRI; however, some CPMs cases are not detected in the prenatal period due to a lack of suspicion and imaging studies[9,11,18,22]. In this analysis, only one patient had a prenatal Doppler ultrasound study that suggested CPM as a diagnostic possibility. This patient underwent surgery at 6 months of age, and the diagnosis of extralobar pulmonary sequestration was confirmed; he had a good clinical and radiologic evolution.

The surgical management of CPMs asymptomatic cases is still poorly defined; some studies propose expectant management and surgical intervention when patients develop symptoms, expecting that late surgery may be safer and reduce complications[19,20,22-24]. In contrast, it is also argued that early intervention at 3 to 6 months could reduce the risk of respiratory infections, hemoptysis, pneumothorax, risk of malignancy and may allow compensatory lung growth[5,20,23,24]. In our study, half of the patients underwent surgery before one year of age, regardless of symptomatology, and showed adequate evolution during hospitalization.

Symptomatic CPM should be managed surgically; lobectomy is the best treatment to avoid the recurrence of pulmonary infection and risk of malignancy[19,20,25,26]. As CT is unreliable in delineating the lung lesion from the healthy zone, subtotal lobectomy or segmentectomy could leave remnants of the lesion if only imaging is relied upon. This consideration is important due to the association between CPAM and the development

of malignancies such as pleuropulmonary blastoma or bronchioalveolar carcinoma[20,26,27]. Surgical management of pulmonary sequestration is also recommended to obtain an anatomopathological diagnosis and avoid malignancy risk; however, endovascular management has been reported as a conservative treatment[28,29]. In these 70 cases, the most frequent surgical procedure was lobectomy, and two patients underwent embolization for pulmonary sequestration.

At present, video-assisted thoracoscopic surgery (VATS) is used by some centers with a complication rate of 12.5% and a conversion rate of 12.5%. This technique reduces the length of hospital stay by up to half and reduces post-operative pain compared to traditional thoracotomy. Therefore, this is the procedure of choice for CPM resection in these centers[22,27,30,31]. In our study, all cases underwent conventional thoracotomy with a median hospital stay of 26 days; no VATS was performed.

There are few studies on post-operative morbidity and mortality of CPMs; factors to be considered are age at the time of surgery, other associated pulmonary pathologies, the extent of disease, hospital resolution capacity, and surgeon learning curve[22,25,31,32,]. In our report, the main complications were pneumonia and pneumothorax. Patients with these complications received medical management with favorable clinical evolution.

Our study has certain limitations: the results cannot be extrapolated to the general population because they are based on data from a single institution, and because it is a national reference hospital, an overestimation of cases could be found. In addition, the description of clinical manifestations may be subject to recall bias by family members (mainly in asymptomatic participants) who provided the information described in the medical records. Despite these limitations, this study contributes to the knowledge and description of a rare pathology in children, such as CPMs, with a considerable number of cases compared to other Latin American studies[5,13,14].

In INSN-Breña, CPMs were more frequent in males, and recurrent bronchopulmonary infections were their main clinical manifestation. The most common types of CPMs were CPAM and pulmonary sequestration. Lobectomy was the most common surgical procedure, and pneumonia was the main complication during hospitalization.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained approval from the Ethics Committee for analysis and publication of routinely acquired clinical data and informed consent was not required for this retrospective observational study.

Conflicts of interest

The authors declare no conflicts of interest.

Funding

No funding.

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