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Clinical Characteristics of Congenital Pulmonary Airway Malformation of The Lungs: A Single-Center Study

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Abstract

Objective: Congenital pulmonary airway malformation (CPAM) is a rare developmental abnormality of the lungs. We investigated clinical characteristics of CPAM based on patient age at diagnosis. *Methods:* In this retrospective study, we analyzed the medical records of 51 patients diagnosed with CPAM at Pusan National University Hospital between January 2000 and December 2019. *Results:* We investigated 39 children and 12 adults. The mean age at the diagnosis of the patients was 15.9 ± 14.3 years. The mean ages of children and adults at the time of diagnosis of CPAM were 6.8 ± 5.8 years and 31.2 ± 10.2 years, respectively. Among the 51 patients investigated, 20 (39%) were asymptomatic and 31 (61%) showed clinical symptoms, such as dry cough, recurrent respiratory infections, and dyspnea at the time of diagnosis. Notably, clinical symptoms at diagnosis were observed in 28 children (72%) and in only 3 adults (25%) ($P = 0.006$), and children were more symptomatic than adults. Children with large cysts tended to be more symptomatic than those with small cysts ($P < 0.001$). Combined anomalies were detected in 12 patients (23%). Patients with cystic lesions in the right lower lobe of the lung showed a higher prevalence of combined anomalies (P = 0.015). Surgical resection was performed in 40 patients (78%), and all patients showed good prognosis. *Conclusion:* This study revealed that the presence of clinical symptoms of CPAM differed between children and adults, depending on the age at diagnosis and that patients with cysts in the right lower lobe of the lungs tended to show a higher prevalence of combined anomalies. (Allergy Asthma Respir Dis 2021, 9: 21–26)

Keywords

Congenital pulmonary airway malformation
Child
Adult

1. Introduction

Congenital pulmonary airway malformation is a rare condition caused by developmental abnormalities of the lower respiratory tract and was formerly referred to as congenital cystic adenomatoid malformation [1]. It is a relatively rare condition with an incidence of 1 per 10,000–35,000 live births, but accounts for 95% of congenital cystic lung lesions [2,3]. The pathogenesis of the disease is unclear, but it has been described as a disruption of fetal bronchial development and airway obstruction, or as an abnormality in lung development caused by abnormal hyperplastic changes in the bronchioles, resulting in multiple lung tissue cystic lesions with alveolar dilation and proliferation of bronchial structures [4,5].

Patients with congenital pulmonary airway malformations may present with symptoms such as respiratory distress or frequent respiratory infections in the neonatal period, but some patients are diagnosed on incidental imaging studies without symptoms [6]. Previous studies have shown that approximately 80–85% of patients are diagnosed during antenatal or neonatal period and rarely in adults [7,8].

To date, most clinical studies of congenital pulmonary airway malformations have focused on the clinical role of prenatal diagnosis of the condition and the assessment of the need for or timing of surgery [9]. Prenatal ultrasonography or fetal magnetic resonance imaging can diagnose congenital pulmonary airway malformations in most fetuses at the gestational age of 18–20 weeks [10,11]. Some patients undergo antenatal interventions such as thoracentesis, thoracocentesis, thoracoamniocentesis, and amniocentesis because fetal death may occur due to hydrops fetalis, hydrothorax, or severe oligohydramnios, and there are studies showing early surgery within the first week of life in neonates who develop symptoms such as severe respiratory distress after birth [12]. However, early planned surgery is sometimes performed in the absence of clinical symptoms after birth, but the appropriate timing of surgery remains controversial [13,14].

Korean studies of congenital pulmonary airway malformations in the last decade have included analyses of children diagnosed prenatally [12–15] and studies of newly diagnosed patients in adulthood [8,16]. However, there is a lack of studies that include all ages of children and adults at the time of diagnosis. Therefore, the authors performed a comparative analysis of the clinical presentation, imaging findings, histological classification, concomitant malformations, treatment methods, and prognosis of a total of 51 pediatric and adult patients diagnosed with congenital pulmonary airway malformations at our institution from 2000 to 2019.

2. Methodology

This study included patients diagnosed with congenital pulmonary airway malformations at Pusan National University Hospital from January 2000 to December 2019. Patients who were suspected of having congenital emphysema on antenatal examination or simple chest X-ray and underwent additional examinations, including chest computed tomography and histological examination, but were not diagnosed with congenital pulmonary airway malformations and did not have at least two follow-up visits were excluded. This study was approved by the Institutional Review Board of Pusan National University Hospital (Institutional Review Board number: H-2003-007-088).

The authors retrospectively analyzed the patients' gender, age at diagnosis, clinical manifestations, lesion location, histological classification, comorbidities, surgical methods and outcomes, and complications through medical records. According to the age at the time of diagnosis of congenital pulmonary airway malformation, patients under 18 years of age were classified as children and adults over 18 years of age. All patients underwent plain chest X-ray and chest computed tomography, and imaging findings were analyzed. Cysts were classified as large if they occupied more than 20% of one lung and as small if they occupied less than 20% [17]. Histological

Table 1. Stocker histopathological classification (2002)

Type 0	Development arrested at the stage of trachea/bronchia formation; tracheal epithelium. Cysts smaller than 0.5 cm; tracheal epithelium; presence of cartilage.
Type 1	Development arrested at the stage of bronchia formation; bronchial epithelium. Large 4–10 cm cysts, cartilage rarely present, squamous-like epithelium.
Type 2	Development of the bronchial tree arrested at the glandular stage. Multiple cysts 0.5–2 cm, covered with columnar epithelium.
Type 3	Development of the bronchial trees arrested at the glandular stage, typical adenomatoid malformation. Multiple cysts < 0.5 cm, covered with columnar epithelium.
Type 4	Development arrested at the stage of bronchia formation; acinar epithelium. Maximum cysts size 7 cm, cartilage absent, association with pleuropulmonary blastoma.

classification for patients who underwent histological examination was based on the Stocker classification (Table 1) [18].

Statistical analysis was performed using IBM SPSS Statistic 23.0 (IBM Co., Armonk, NY, USA), and categorical variables were described as number of patients and percentages. Variables for the association of lesion location with comorbidity, and differences in the presence or absence of symptoms according to lesion size were analyzed using the Fisher exact test, and a *P* value of less than 0.05 was considered statistically significant.

3. Results

3.1. Gender and age distribution

From January 2000 to December 2019, 64 patients were diagnosed with congenital pulmonary airway malformations. Among them, 13 patients who were not observed for at least two follow-up visits were excluded, leaving a total number 51 study subjects, with 33 males (65%) and 18 females (35%) (Table 2).

When classified by age at the time of diagnosis, there were 39 pediatric patients and 12 adult patients. Among these, the largest group consisted of patients under 1 year of age, including 19 cases diagnosed during prenatal examinations, totaling 20 patients

Table 2. Characteristic features of congenital pulmonary airway malformation (*n* = 51)

Characteristic	Value
Age at diagnosis (yr)	
Mean ± SD (*n* = 32)	15.9 ± 14.3
Range	0–50
< 1	20 (39)
1–10	11 (21)
10–18	8 (16)
18–30	7 (14)
30–40	3 (6)
> 40	2 (4)
Sex	
Male	33 (65)
Female	18 (35)
Symptoms or clinical presentation at diagnosis	
Prenatally diagnosed	19 (37)
Asymptomatic	20 (39)
Nonproductive cough	12 (24)
Pulmonary infection	7 (14)
Dyspnea	5 (10)
Chest pain	4 (8)
Tachypnea	3 (6)
Stocker classification (*n* = 30)	
0	0 (0)
1	21 (70)
2	8 (27)
3	1 (3)
4	0 (0)
Radiologic findings	
Multiple cysts	38 (74)
Solitary cysts	13 (26)
Combined anomalies	
None	39 (76)
Pulmonary sequestration	7 (14)
Emphysema	2 (4)
Bronchogenic cysts	1 (2)
Patent ductus arteriosus	1 (2)
Accessory bronchus	1 (2)
Treatment	
Lobectomy	33 (65)
Transfer or plan to operation	11 (22)
Wedge resection	5 (10)
Pneumonectomy	2 (3)

Note: Values are presented as number of patients (%) unless otherwise indicated.

(39%). The average age at the time of diagnosis for patients excluding those diagnosed during prenatal examinations was 15.9 ± 14.3 years (Table 2). When categorized into pediatric and adult groups based on their age at diagnosis, the average ages were found to be 6.8 ± 5.8 years and 31.2 ± 10.2 years, respectively.

3.2. Clinical presentation

Among the 51 patients, 19 individuals (37%) were born at the hospital with suspected congenital tracheal airway anomalies identified during prenatal examinations or were transferred to the hospital shortly after birth from other medical facilities for further evaluation. Among these, 9 out of 19 patients (47%) presented clinical symptoms such as dyspnea, tachypnoea, and rib cage depression. A total of 20 patients (39%) were later confirmed to have congenital pulmonary airway malformations from incidental tests such as medical examinations without clinical symptoms, including 10 patients who were suspected of having the condition during prenatal examinations but had no symptoms after birth. At the time of diagnosis, the clinical symptoms observed were as follows: dry cough in 12 patients (24%), recurrent respiratory infections in 7 patients (14%), shortness of breath in 5 patients (10%), chest pain in 4 patients (8%), and tachypnoea in 3 patients (6%) (Table 2). Comparing the proportion of children and adults with clinical symptoms at diagnosis, 28 out of 39 children (72%) had symptoms compared to only 3 out of 12 adults (25%), with a statistically significant higher proportion of children with clinical symptoms than adults (P = 0.006) (Table 3).

3.3. Imaging and histological findings

Analysis of the chest computed tomography of all patients showed that 38 (74%) had multiple cystic lesions and 13 (26%) had single cystic lesions (Table 2). The lesions were most commonly located in the right lower lobe in 17 patients (33%), followed by the left lower lobe in 14 patients (27%), the right upper lobe

Table 3. Comparison of characteristics between children and adults

Characteristic	Children (n = 39)	Adults (n = 12)	P-value*
Symptom at diagnosis			0.006
None	11 (28)	9 (75)	
Present	28 (72)	3 (25)	
Locations (only single lobe, n = 48)			
RUL	4 (11)	5 (42)	0.032
RML	2 (6)	0 (0)	1.000
RLL	12 (33)	5 (42)	0.730
LUL	4 (11)	2 (16)	0.631
LLL	14 (39)	0 (0)	0.010
Cysts			0.271
Small lesion	9 (23)	5 (41)	
Large lesion	30 (77)	7 (59)	
Stocker classification (n = 30)			1.000
1	16 (69)	5 (72)	
2	6 (27)	2 (28)	
3	1 (4)	0 (0)	
Combined anomaly			1.000
None	30 (90)	9 (75)	
Present	9 (10)	3 (25)	

Note: Values are presented as number (%); R, right; L, left; UL, upper lobe; ML, middle lobe; LL, lower lobe; *Fisher exact test was done.

in 9 patients (18%), the left upper lobe in 6 patients (12%), and the right middle lobe in 2 patients (4%). Three cases (6%) involved multiple lobes. In children, cysts were most common in the left lower lobe with 14 (39%) (P = 0.010). In adults, there were 5 (42%) in the right upper and right lower lobes, with significantly more lesions in the right upper lobe in adults compared to children (P = 0.032) (Table 3).

The mean size of the lesions was 3.9 cm×2.9 cm in children and 5.6 cm×4.1 cm in adults. Large lesions occupying more than 20% of the lung area were found in 30 (77%) children and 7 (59%) adults (Table 3). In addition, in all patients, clinical symptoms were significantly more common with larger cyst size (P < 0.001), and when analyzed separately by age at diagnosis, clinical symptoms were more common with larger cyst size in children (P < 0.001), but no significant relationship between symptoms and size was found in adults (P = 0.204) (Table 4).

A total of 30 of 51 patients (59%) underwent histological examination, with type 1 predominating in 21 patients (70%) according to the Stocker classification. Type 2 was identified in 8 patients (27%) and type 3 in 1 patient (3%). No patients were diagnosed with types 0 and 4 (Table 2).

3.4. Comorbidities

Comorbidities were identified in 12 of the 51 patients (24%), including pulmonary sequestration in 7 patients, emphysema in 2 patients, bronchogenic cyst in 1 patient, patent ductus arteriosus in 1 patient, and accessory bronchus in 1 (Table 2). There were no other renal, digestive, or other comorbidities, and there was no difference in the presence of comorbid anomalies between children and adults. Among the 48 patients with unilateral lung lesions, excluding the 3 patients with lesions involving multiple lobes, the location of the lung lesion and the presence of associated comorbidities were examined. The results showed a significant association between the presence of comorbidities and lesions in the right lower lobe (P = 0.015) (Table 5).

3.5. Treatment and prognosis

Surgical resection was performed in 40 of the 51 patients (78%). Of those who underwent surgical treatment, the majority–33 patients (65%), underwent lobectomy, 5 patients (10%) underwent wedge resection, and 2 patients (3%) underwent pneumonectomy. The mean age of the 40 patients

Table 4. Association of cyst size and symptoms

	Total (n = 51)		Children (n = 39)		Adults (n = 12)	
	Small	Large	Small	Large	Small	Large
Symptom (-)	12	8	7	4	5	4
Symptom (+)	2	29	2	26	0	3
P-value*	< 0.001		< 0.001		0.204	

Note: *Fisher exact test was done.

Table 5. Association of location of CPAM and combined anomalies

Location	Without combined anomaly(n = 36)	With combined anomaly(n = 12)	*P*-value*
Right upper lobe	7(19)	2(17)	1.000
Right middle lobe	2(6)	0(0)	1.000
Right lower lobe	9(25)	8(67)	0.015
Left upper lobe	5(14)	1(8)	1.000
Left lower lobe	13(36)	1(8)	0.081

Note: Values are presented as number (%); *Fisher exact test was done.

who underwent surgery was 11.7 ± 13.8 years (range: 0–50 years). All patients who underwent surgery had a favorable prognosis and had no complications (Table 2).

Out of the 11 patients who did not undergo surgical treatment, 6 patients were transferred to other hospitals for surgical treatment. The remaining 5 patients who did not undergo surgery were managed with regular follow-ups in the outpatient department. The average age of these 5 patients was 1.6 ± 2.2 years (range: 0–6 years). Patients are currently asymptomatic and are under outpatient observation with the intention of performing surgery at a later date in the event of clinical symptoms or recurrent respiratory infections. The average follow-up period for these patients was 22.4 months.

4. Discussion

This study is significant as it compared the clinical characteristics of pediatric and adult patients with congenital tracheal airway anomalies based on the age at the time of diagnosis. The results showed that pediatric patients more frequently presented clinical symptoms at the time of diagnosis compared to adults. Additionally, the study found that the risk of concomitant anomalies is higher when the congenital pulmonary airway anomaly is located in the right lower lobe of the lung.

Congenital pulmonary airway anomalies are reported to be more common in males, although the cause is unclear [19], and this was confirmed in our study, with 65% of patients were male and 35% of patients were female. Additionally, the study revealed a significant difference in the clinical symptoms between pediatric and adult patients at the time of diagnosis, with 72% of pediatric patients presenting symptoms compared to 25% of adult patients ($P = 0.006$). Among these children with clinical symptoms, 77% had large lesions occupying more than 20% of one lung. The relationship between the size of the lesion and the presence of symptoms showed statistical significance in pediatric patients, but not in adult patients. Previous studies have also reported that large cysts in neonates compress the normal lung tissues, causing symptoms such as respiratory distress [12]. Therefore, in children, the larger the size of the lesion in relation to the total lung area, the more likely it is that clinical symptoms will occur, suggesting that congenital pulmonary airway malformations are diagnosed at an earlier age.

In general, due to the development of prenatal ultrasound, very small-sized lesions diagnosed prenatally tend to be asymptomatic at birth, and it is known that about 25% of newborns diagnosed prenatally develop symptoms [1,20,21]. However, in this study, 47% of newborns diagnosed prenatally developed clinical symptoms such as dyspnea, tachypnoea, and rib cage depression after birth, which is a higher percentage than previous studies. This may be due to the inclusion of more severe patients in the university hospital. Previous studies have reported that adult patients with symptoms mainly have lesions in the lower lobe [14]. However, in this study, out of 12 adult patients diagnosed, 3 patients showed symptoms, with 2 cases (67%) involving the upper lobe and 1 case (33%) involving the lower lobe. Therefore, further studies on the size, location, and clinical manifestations of lesions in larger sample size of adult patients are warranted.

It has been reported that approximately 15–20% of patients with congenital pulmonary airway anomalies have comorbidities, usually in the heart or kidneys [4,22]. In this study, 24% of patients had other anomalies, with pulmonary sequestration being the most common comorbidity at 14%. Another interesting finding in this study was that lesions in the right lower lobe were significantly more likely to be accompanied by other anomalies ($P = 0.015$). To the authors' knowledge, there have been no reports on the relationship between the location of congenital pulmonary airway anomaly lesions and comorbid anomalies. Therefore, further research is needed to determine whether additional testing is needed to identify comorbidities in patients

with lesions in the right lower lobe.

In 1977, pathologist Stocker proposed a histological classification of congenital pulmonary airway malformations into three types: types 1, 2, and 3 [23]. In 2002, he added types 0 and 4 to the previously published types and now classifies them into five types (Table 1). Previous studies have shown that more than 65% of congenital pulmonary airway malformations are type 1, 20–25% are type 2, and 5–10% are type 3 and 4, with type 0 being the rarest form, with a distribution of approximately 1–3%.18 Based on the histopathological examinations performed in this study's patients, it was reconfirmed that type 1 accounted for 70%, type 2 for 27%, and type 3 for 3%, reaffirming that type 1 and type 2 are the most prevalent types.

The treatment of congenital pulmonary airway anomalies is surgical resection in the presence of clinical symptoms such as recurrent respiratory tract infections and severe dyspnea. Surgical resection is performed in cases with recurrent infections, clinical symptoms, and risk of malignant progression, as well as in cases diagnosed in adulthood and requiring differentiation from malignancy [1,24]. However, there have been reports of spontaneous resolution of the lesions without any treatment [25], and there is still ongoing debate regarding the need for surgical resection in asymptomatic patients [26,27]. In this study, 40 of the 51 patients identified underwent surgical resection. The patients who underwent surgical treatment at our institution for symptoms such as recurrent respiratory infections or for differentiation from malignancy in adults had good prognosis without postoperative sequelae.

There are several limitations to this study. This was a retrospective study analyzing only medical records from a single institutional university hospital, and the sample size was small. Because it was based on medical records, some data may have been incomplete or missing.

However, this study analyzed the clinical presentation of congenital pulmonary airway anomalies and showed that there were differences in the presence or absence of clinical symptoms at diagnosis when children and adults were divided into groups based on age at diagnosis. It is also important to note that this study suggests that congenital pulmonary airway anomalies located in the right lower lobe are more likely to be accompanied by other anomalies. This finding adds new insights to the understanding of these anomalies.

Disclosure statement

The authors declare no conflict of interest.

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