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Evaluation of Prenatal, Perinatal and Postnatal Outcomes and Imaging Modalities of Congenital Lung Malformations Diagnosed Prenatally: A Five-year Analysis in a Tertiary Center

Prenatal Tanı Alan Konjenital Akciğer Malformasyonlarının Antenatal, Perinatal ve Postnatal Sonuçlarının ve Görüntüleme Yöntemlerinin Değerlendirilmesi: Tersiyer Bir Merkezde Beş Yıllık Analiz

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ABSTRACT

Objective: To evaluate the antenatal, perinatal, and postnatal outcomes of congenital lung malformations (CLM) diagnosed prenatally.

Methods: This was a retrospective cross-sectional study evaluating prenatally diagnosed CLMs. Prenatal findings such as diagnosis weeks, additional anomalies, karyotype results, and pregnancy outcomes of the cases were collected from the hospital digital record system. The data of the postnatal long-term course and outcomes of the cases were obtained by contacting the parents by telephone.

Results: Forty-seven CLM cases were included in the study. 12.8% of the cases had structural anomalies. Chromosome analysis was performed on 8 (17%) cases and all of them had a normal karyotype. Forty-four (93.7%) cases had a live birth. 19.1% of the cases gave birth prematurely. The newborns had low APGAR scores (<7) at the 1st minute in 25.5% of the cases and at the 5th min in 12.8% of the cases. Respiratory distress syndrome (RDS) was present in 28.9% of the newborns, and 15.6% of the cases were hospitalized in the neonatal intensive care unit (NICU). Remission was achieved in 20 (48.8%) cases in the postnatal follow-up. Eight (19.5%) cases persisted, and 13 (31.7%) of all cases were operated. The compatibility of prenatal ultrasonography and magnetic resonance imaging results with lesion types was 70.2%.

Conclusion: While CLM cases mostly showed a favorable prognosis in the antenatal period, adverse outcomes such as RDS and NICU admission were high in the perinatal period. Surgical resection was required in approximately 1/3 of the cases in the postnatal period.

Keywords: Bronchopulmonary sequestration, congenital pulmonary airway malformation, congenital lung malformations, pregnancy outcomes, postnatal outcomes

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ÖZ

Amaç: Prenatal tanı alan konjenital akciğer malformasyonu (KAM) olgularının antenatal, perinatal ve postnatal sonuçlarının değerlendirilmesidir.

Yöntem: Bu retrospektif kesitsel çalışmada prenatal tanı alan KAM olguları değerlendirildi. Olguların tanı haftaları, ek anomalileri, karyotip sonuçları, gebelik sonuçları gibi prenatal bulguları hastane dijital kayıt sisteminden elde edildi. Postnatal uzun dönem sonuçlarına ait veriler katılımcılara telefon ile ulaşılarak elde edildi.

Bulgular: Çalışmaya 47 KAM olgusu dahil edildi. Olguların %12,8'inde yapısal anomaliler vardı. Sekiz (%17) olguya kromozom analizi yapıldı ve hepsinin karyotipi normaldi. Olguların 44'ü (%93,7) canlı doğum yaptı. Olguların %19,1'i preterm doğumdu. Yenidoğanların %25,5'inde 1. dakikada, %12,8'inde 5. dakikada <7 APGAR skoru mevcuttu. Yenidoğanların %28,9'unda respiratuar distres sendromu (RDS) mevcuttu ve olguların %15,6'sı yenidoğan yoğun bakım ünitesine (YYBÜ) yatırıldı. Postnatal takipte 20 (%48,8) olguda remisyon izlendi. Olguların 8'i (%19,5) sebat etti ve tüm olguların 13'ü (%31,7) opere edildi. Prenatal ultrasonografi ve manyetik rezonans görüntüleme sonuçlarının lezyon tipleri ile uyumluluğu %70,2 idi.

Sonuç: KAM olguları antenatal dönemde çoğunlukla olumlu prognoz gösterirken, perinatal dönemde RDS ve YYBÜ'ye yatış gibi olumsuz sonuçlar yüksekti. Postnatal dönemde olguların yaklaşık 1/3'ünde cerrahi rezeksiyon gerekmiştir.

Anahtar Kelimeler: Bronkopulmoner sekestrasyon, konjenital pulmoner hava yolu, konjenital akciğer malformasyonları, gebelik sonuçları, doğum sonrası sonuçlar

INTRODUCTION

Congenital lung malformations (CLM) are a heterogeneous group encompassing a continuum of developmental disorders that include the pulmonary parenchyma, bronchi, arterial supply, and venous drainage. It includes abnormal connections of one or more of the four major components of lung tissue: tracheobronchial tree, lung parenchyma, arterial supply, and venous drainage.[1] According to the involved components of the lung, it includes congenital pulmonary airway malformation (CPAM), bronchopulmonary sequestration (BPS), hybrid lesions containing elements of both CPAM and BPS, bronchial atresia, congenital high airway obstruction syndrome (CHAOS), and bronchogenic cysts.[2] The pathogenesis of these malformations is not fully understood. According to the proposed theories, these lesions are thought to result from defective foregut budding or pulmonary dysplastic changes secondary to airway obstruction. Hybrid or overlapping lesions may be explained by variability in the timing and severity of airway obstruction.[3-5]

CLMs are rare, and their prenatally estimated incidence is about 1 in 2400 live births.[6] Prenatal diagnosis of CLM has increased significantly recently due to the more widespread and improved prenatal ultrasonography (US) screening. In most cases, a well-done US provides the correct diagnosis, but in selected cases, supplemental magnetic resonance imaging (MRI) is increasingly applied for additional useful information. However, prenatal imaging should not attempt to make a histological diagnosis. Although these malformations have overlapping features, hybrid lesions may also contain components of two distinct types of malformations.[7,8]

Prenatal findings are useful in predicting the antenatal prognosis as well as determining the type of lung malformation. Intrauterine indicators of poor prognosis include large lesions, bilateral lung involvement, and hydrops.[5,9] Fetal hydrops due to CLM is associated with a high rate of intrauterine fetal loss and intervention or preterm delivery is indicated.[10] At birth, most CLMs have a favorable prognosis. Delivery is usually uncomplicated. Most neonates (>75%) are asymptomatic, and only a minority require respiratory support.[11] CLM is mostly asymptomatic in the postnatal period, but undiagnosed or untreated CLM can cause long-term consequences such as recurrent pulmonary infection, pneumothorax and malignancy.[12]

In this study, we evaluated the antenatal, perinatal, and postnatal outcomes of CLM diagnosed prenatally in a tertiary center.

METHODS

This retrospective cross-sectional study was conducted with pregnant women diagnosed with CLM prenatally, who were followed up in the Department of Perinatology, University of Health Sciences Turkey, İzmir Tepecik Training and Research Hospital İzmir, Turkey, between January 2017 and December 2021. The study protocol was approved by the University of Health Sciences Turkey, İzmir Tepecik Training and Research Hospital Local Institutional Ethics Committee (approval number: 2022/03-09, date: 15.03.2022).

Pregnant women who were diagnosed with fetal CLM by US and/or MRI during the prenatal period and delivered at our hospital between the relevant dates were included in the study. Antenatal and perinatal outcomes were collected from the hospital digital record system. The data of the postnatal long-term course and outcomes of the cases were obtained by contacting the parents by telephone. Pregnant women whose data regarding their antenatal or perinatal outcomes could not be reached were excluded from the study.

The definition developed by Stocker et al.[13] was used in the diagnosis and classification of CPAM. Lesions with single or multiple large cysts (3-10 cm in diameter) were defined as CPAM type 1, lesions containing smaller cysts

(0.5-2 cm) as CPAM type 2, and solid lesions with cysts smaller than 0.2 cm were defined as CPAM type 3 (Figure 1). BPS is located in the lower lobes of the lung and is mostly on the left. The imaging findings of BPS usually include a solid, well-defined, uniform, and hyperintense mass. The differential diagnosis includes CPAM type 3, and systemic vascular supply should be demonstrated (Figure 2). Additionally, extralobar BPS should be differentiated from neuroblastoma and adrenal hemorrhage.[14] CHAOS imaging typically shows enlarged hyperintense lungs and a flattened or outward-facing hemidiaphragm. Imaging with MRI can help locate the obstruction by showing a dilated airway.[15] Bronchial atresia most commonly occurs in the apical and posterior segments of the left upper lobe and presents as a focal thoracic mass of homogeneous density.[14]

Prenatal findings such as diagnosis weeks, additional anomalies, karyotype results and pregnancy, perinatal and postnatal long-term outcomes of all cases were evaluated. The association between prenatal US and MRI imaging results of the cases was also evaluated.

Statistical Analysis

Data are represented as mean±standard deviation or n (%). Microsoft Excel Professional Plus 2019 and IBM® Statistical Package for the Social Sciences Statistics v26.0 were used to analyze the data.

RESULTS

A total of 47 CLM cases diagnosed prenatally between January 2017 and December 2021 were included in the study. The mean maternal age of the cases was 28.4±4.6 (Table 1). The pregnancy findings and pregnancy outcomes of the cases are presented in Table 2. There was a one-week difference between prenatal US imaging time and MRI imaging time, with a mean US week of 22±3 and a mean MRI week of 23±3. Additional findings were as follows: 10.6% of the cases had soft markers (intracardiac echogenic focus and pelviectasis) and 12.8% had structural anomalies [fetal abdominal ascites, hypertelorism, ventriculomegaly and ventricular septal defect (VSD)]. No additional findings were found in 76.6% of the cases. Chromosome analysis was performed on 8 (17%) cases, and all of them had a normal karyotype. The pregnancy outcomes of the cases were as follows: forty-four (93.7%) cases had live births, one

Table 1. Maternal demographic and medical characteristics

	Mean±SD or n, (%)
Maternal age (year) (mean±SD)	28.4±4.6
Adolescent pregnancy ≤19 year (n, %)	0
Advanced age pregnancy ≥35 year (n,%)	2 (4.3%)
Parity (n, %)	
Nulliparous	19 (40.4%)
Multiparous	28 (59.6%)

SD: Standard deviation

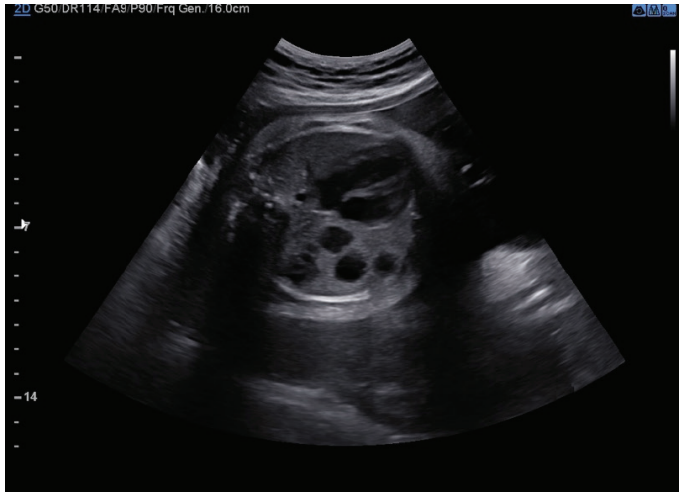

Figure 1. Ultrasonographic image of congenital pulmonary airway malformation type 1 located in the fetal right lung

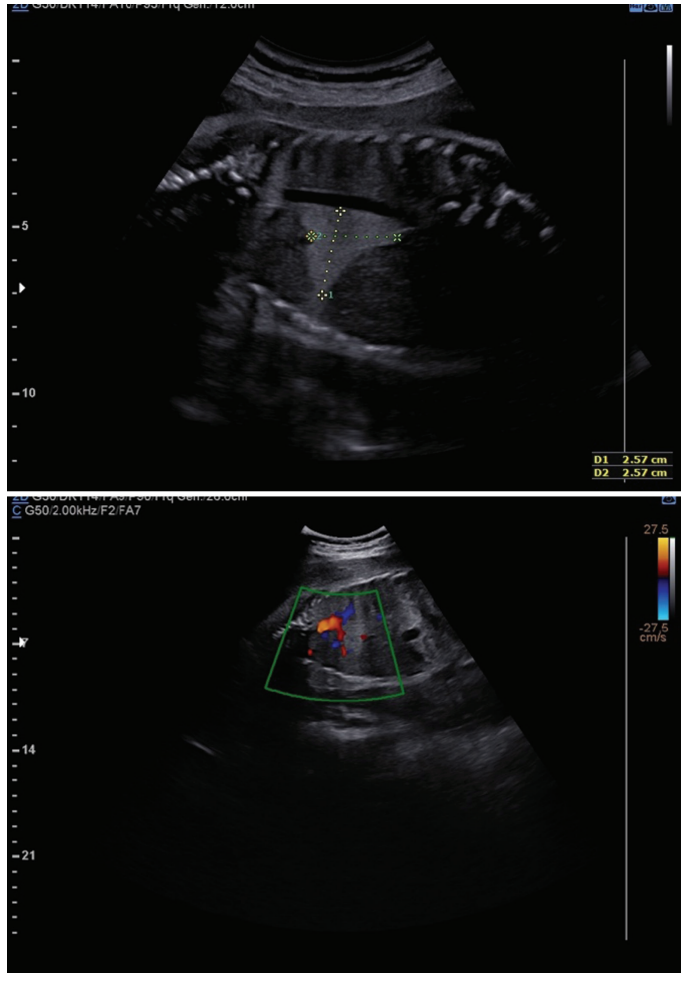

Figure 2. Ultrasonographic image of bronchopulmonary sequestration and its systemic vascular supply

BPS case was terminated due to hydrops fetalis, one CPAM type 1 case had stillbirth, and one CHAOS case had early neonatal death.

Perinatal and postnatal outcomes of the cases are presented in Table 3. The mean delivery week of the cases was 38±3. 19.1% of the cases gave birth prematurely. 10.6% of newborns were low birth weight (<2500 g). The newborns had low APGAR scores (<7) at the 1st minute in 25.5% of the cases and at the 5th min in 12.8% of the cases. Respiratory distress syndrome (RDS) was present in 28.9% of the newborns, and 15.6% of the cases were admitted to the neonatal intensive care unit (NICU). Postnatal outcomes included forty-one newborns. Postnatal outcomes of antenatally diagnosed one CPAM type 3 case and two BPS cases with could not be reached. Remission was achieved in 20 (48.8%) cases in the postnatal follow-up. Eight (19.5%) cases persisted, and 13 (31.7%) of all cases were operated.

Prenatal diagnostic comparisons of the cases with US and MRI are presented in Table 4. In prenatal imaging, US diagnoses were compatible with MRI diagnoses in 85.7% of CPAM type 1 and 81.3% of CPAM type 2. However, in MRI evaluations of cases diagnosed with CPAM type 3 by US, BPS was found in 46.7% and bronchial atresia in 13.3%. 87.5% of the cases diagnosed with BPS by US were confirmed by MRI. There was a case of CHAOS, and the compatibility between imaging modalities was 100%. The compatibility of prenatal US and MRI results according to the types of lesions is also presented in Table 5.

DISCUSSION

CLM represents a heterogeneous group of abnormalities that affect the developmental stages of the fetal lung. Although its etiology has not been fully elucidated, several hypotheses have been proposed. While both type and histopathology are associated with the timing of embryological damage, they are presumed to be due to anomalies of airway embryogenesis.[16,17] However, a predisposing factor that can be associated with CLM has not yet been suggested among the reasons shown to be associated with other congenital anomalies such as genetic mutation, family history, and exposure to teratogen that may cause this developmental disorder of the fetal lung.[12] The number of prenatally diagnosed cases has increased over the years. Stocker et al.[6] retrospectively evaluated 133 CLM cases and showed that the mean incidence of CLM increased from 1.27 per 10.000 births between 1994-1998 and gradually increased to 4.15 per 10.000 births between 2008 and 2012. They also showed that prenatal findings were detected in the

Table 2. Antenatal findings and pregnancy outcomes of the cases

	CPAM type 1 n=7	CPAM type 2 n=14	CPAM type 3 n=7	BPS n=15	CHAOS n=1	Bronchial atresia n=3	Total
US gestational week (mean±SD)	22±5	22±2	22±3	22±2	21	23±3	22±3
MRI gestational week (mean±SD)	23±4	23±2	23±3	23±2	22	26±4	23±3
MRI additional findings (n, %)							
Acid	0	0	0	1 (6.7%)	1 (100%)	0	2 (4.3%)
Hypertelorism	0	0	1 (14.3%)	0	0	0	1 (2.1%)
Intracardiac echogenic focus	0	1 (7.1%)	0	0	0	0	1 (2.1%)
Pelviectasis	1 (14.3%)	2 (14.3%)	1 (14.3%)	0	0	0	4 (8.5%)
Ventriculomegaly	1 (7.1%)	0	0	1 (6.7%)	0	0	2 (4.3%)
Ventricular septal defect	0	0	0	1 (6.7%)	0	0	1 (2.1%)
None	5	11	5	12	0	3	36 (76.6%)
Chromosome analysis (n, %)							
Normal karyotype	3 (42.9%)	1 (7.1%)	2 (28.6%)	1 (6.7%)	0	1 (33.3%)	8 (17%)
The patient refused/unknown/ unable to follow	4 (57.1%)	13 (92.9%)	5 (71.4%)	14 (93.3%)	1 (100%)	2 (66.7%)	39 (83%)
Pregnancy outcome							
Termination	0	0	0	1 (6.7%)	0	0	1 (2.1%)
Live birth	6 (85.7%)	14 (100%)	7 (100%)	14 (83.3%)	0	3 (100%)	44 (93.7%)
Stillbirth	1 (14.3%)	0	0	0	0	0	1 (2.1%)
Neonatal death	0	0	0	0	1 (100%)	0	1 (2.1%)

BPS: Bronchopulmonary sequestration, CHAOS: Congenital high airway obstruction syndrome, CPAM: Congenital pulmonary airway malformation, MRI: Magnetic resonance imaging, SD: Standard deviation, US: Ultrasonography

early second trimester (21.1 weeks) on US, similar to our results (22±3 weeks).

Abnormalities associated with CLM are unusual but are more common with type 2. They are usually isolated anomalies and secondary findings related to cardiac failure such as ascites, hydrops and pleural effusion are detected secondary to the fetal lesion affecting the cardiac circulation.[18] Our findings also showed that fetal acid was present in two cases with BPS and CHAOS. One CPAM type 1 and one BPS case had mild ventriculomegaly (<12 mm). One CPAM type 3 case had a prenatal finding of hypertelorism, but there was no additional anomaly suggesting a genetic syndrome. One BPS case also had VSD. However, apart from these findings and soft markers, there were no additional major structural anomaly in any case. Additionally, prenatal chromosome analysis was performed in 17% of the cases and karyotype anomaly was not found in any of them.

Although many CLMs have an antenatally favorable prognosis, a fetal risk may occur secondary to the

Table 3. Perinatal and postnatal long-term outcomes of the cases

	CPAM type 1 n=7	CPAM type 2 n=14	CPAM type 3 n=7	BPS n=15	CHAOS n=1	Bronchial atresia n=3	Total
Delivery week (week) (mean±SD)	36.7±4.8	38.3±1.5	39±1.4	37.6±4	37.2	36.6±0.3	38±3
Preterm delivery prevalence (<37 week) (n, %)	1 (14.3%)	3 (21.4%)	1 (14.3%)	2 (13.3%)	0	2 (66.7%)	9 (19.1%)
Fetal gender (n, %)							
Male	5 (71.4%)	7 (50%)	1 (14.3%)	8 (53.3%)	1 (100%)	3 (100%)	25 (53.2%)
Female	2 (28.6%)	7 (50%)	6 (85.7%)	7 (46.7%)	0	0	22 (46.8%)
Birth weight (g)	2769.9±849	3169±302	2950±547	2958±779	2495	3433±375	3012±621
LBW (<2500 g) (n, %)	1 (14.3%)	1 (7.1%)	1 (14.3%)	1 (6.7%)	1 (100%)	0	5 (10.6%)
Macrosomia (≥4000 g) (n, %)	0	0	0	1 (6.7%)	0	0	1 (2.1%)
APGAR score (n, %)							
<7 at 1st minute	3 (42.9%)	4 (28.6%)	2 (28.6%)	2 (13.3%)	1 (100%)	0	12 (25.5%)
<7 at 5th minute	1 (14.3%)	2 (14.3%)	1 (14.3%)	1 (6.7%)	1 (100%)	0	6 (12.8%)
NICU admission (n, %)[a]	0	3 (21.4%)	3 (42.9%)	1 (7.1%)	0	0	7 (15.6%)
RDS (n, %)[a]	3 (50%)	6 (42.9%)	3 (42.9%)	1 (7.1%)	0	0	13 (28.9%)
Postnatal outcomes[b]							
Remission	1 (16.7%)	6 (42.9%)	4 (66.7%)	6 (50%)	0	3 (100%)	20 (48.8%)
Persistence	0	1 (7.1%)	2 (33.3%)	5 (41.7%)	0	0	8 (19.5%)
Operation	5 (83.3%)	7 (50%)	0	1 (8.3%)	0	0	13 (31.7%)

[a]Excludes 1 case of CPAM type 1 and 1 case of BPS, includes a total of 45 live births.

[b]Includes 41 newborns (1 case of CPAM type 3, 2 case of BPS newborns unknown/unable to follow).

BPS: Bronchopulmonary sequestration, CHAOS: Congenital high airway obstruction syndrome, CPAM: Congenital pulmonary airway malformation, LBW: Low birth weight, NICU: Neonatal intensive care unit, RDS: Respiratory distress syndrome, SD: Standard deviation

Table 4. Comparison of prenatal US and MRI diagnoses of the cases

	CPAM type 1	CPAM type 2	CPAM type 3	BPS	CHAOS	Bronchial atresia	Total
CPAM type 1 (n, %)	6 (85.7%)	1 (14.3%)	0	0	0	0	7 (100%)
CPAM type 2 (n, %)	1 (6.3%)	13 (81.3%)	0	1 (6.3%)	0	1 (6.3%)	16 (100%)
CPAM type 3 (n, %)	0	0	6 (40%)	7 (46.7%)	0	2 (13.3%)	15 (100%)
BPS (n, %)	0	0	1 (12.5%)	7 (87.5%)	0	0	8 (100%)
CHAOS (n, %)	0	0	0	0	1 (100%)	0	1 (100%)
Total (n, %)	7 (14.9%)	14 (29.8%)	7 (14.9%)	15 (31.9%)	1 (2.1%)	3 (6.4%)	47 (100%)

BPS: Bronchopulmonary sequestration, CHAOS: Congenital high airway obstruction syndrome, CPAM: Congenital pulmonary airway malformation, MRI: Magnetic resonance imaging, US: Ultrasonography

growth of the mass in some cases. However, there are no universally accepted clinical recommendations or practice guidelines for antenatal management yet. It is difficult to predict the natural course of CLMs during the antenatal period. Although many will enter the proliferative phase in the second trimester, a significant number of prenatal lesions are stable or resolve later in pregnancy.[10] Adzick evaluated more than 600 cases of CLM and reported a significant regression rate of 68% in BPS cases before delivery, compared to 15% in CPAM cases. However, most remained detectable with a postnatal computed tomography (CT) scan.[7] Cavoretto et al.[19] similarly showed 50% antenatal regression in BPS and also reported a similar regression rate for non-hydropic echogenic CPAM. Similarly, our findings showed that CLMs progressed favourable in the antenatal period. Hydrops fetalis developed in only one CPAM type 1 case and one BPS case, and the early BPS case was terminated and the CPAM type 1 case was stillbirth. However, as a limitation of our study, although our data include the antenatal outcomes of the cases, data on the course of the size of the lesions were not included.

The perinatal prognosis of CLM varies according to the lesion type, location, and size. Most cases are asymptomatic at birth. However, 1% neonatal death was reported in BPS and CPAM cases, and emergency surgical resection was reported in 7% of cases. The morbidity is much higher in CHAOS cases.[20,21] Our perinatal findings showed that APGAR scores were <7 at the 1st minute in 25.5% of the cases and the 5th minute in 12.8% of the cases. Additionally, 28.9% of the cases had RDS and 15.6% had NICU admission. Our results showed that CLM adversely affects early neonatal outcomes. Moreover, there is no indication for preterm delivery in CLM cases, except for fetal hydrops.[10] Our perinatal findings demonstrate that the mean week of delivery in CLM cases was 38±3. 19.1% of the cases gave birth prematurely and it was higher than the preterm delivery rates reported as 10% worldwide.[22] More studies are needed to evaluate the association of preterm delivery other than hydrops in CLM cases.

In the postnatal evaluation, it is thought that CLMs diagnosed in the antenatal period often regress or disappear after birth. However, most of the time, adequate imaging methods are not used in the postnatal period. Simple chest radiographs are inadequate. MRI may be helpful, but the gold standard imaging modality is contrast-enhanced CT scanning.[23,24] Our results showed that 48.8% of the cases were in remission in the evaluations extending to the postnatal 5-year follow-up. However, there is no study in the literature showing that CLMs with cystic components can undergo remission with adequate postnatal imaging.[10] The high remission rate of our cases may be related to the fact that most of them were followed by imaging methods such as simple radiography with low benefit, and very few cases were evaluated with CT.

There is no consensus regarding the postnatal management of CLMs. Surgical resection is a common approach for children with symptomatic CLM, but the necessity and timing of surgery in children with asymptomatic CLM remains controversial. The risk of infection for CLMs is between 10% and 30% in the first year of life.[24,25] Some authors have also recommended early surgical resection to take advantage of compensatory lung enlargement.[24,26] It is also known that CLMs are associated with malignancies such as pleuropulmonary blastoma and bronchioloalveolar carcinoma. Although the risk of malignancy is sufficient justification for surgical resection in asymptomatic cases, some authors have reported cases that also developed malignancy after surgical resection.[27] 31.7% of our cases, which included postnatal 5-year follow-up, were operated. CPAM type 1 and CPAM type 2 constituted the majority of the operated cases. These results may be related to the presence of persistent cystic lesions in the clinicians' preference for surgery.

In our study, we also evaluated the association between prenatal imaging methods. The authors recommend prenatal US imaging as the primary imaging modality for fetal evaluation, largely because it is accessible and inexpensive. However, in the presence of inconclusive

Table 5. Compatibility of prenatal US and MRI results according to CLM types

	US and MRI same diagnosis	MRI changes diagnosis	Total
CPAM type 1 (n, %)	6 (85.7%)	1 (14.3%)	7 (100%)
CPAM type 2 (n, %)	13 (81.3%)	3 (18.7%)	16 (100%)
CPAM type 3 (n, %)	6 (40%)	9 (60%)	15 (100%)
BPS (n, %)	7 (87.5%)	1 (12.5%)	8 (100%)
CHAOS (n, %)	1 (100%)	0	1 (100%)
Total (n, %)	33 (70.2%)	14 (29.8%)	496 (100%)

BPS: Bronchopulmonary sequestration, CHAOS: Congenital high airway obstruction syndrome, CLM: Congenital lung malformation, CPAM: Congenital pulmonary airway malformation, MRI: Magnetic resonance imaging, US: Ultrasonography

sonographic findings, the use of fetal MRI imaging is helpful in differential diagnosis.[28,29] Our results showed that 70.2% of prenatal US and MRI scans were compatible. In prenatal US imaging, CPAM types 2 and 3 were the lesions most different from MRI imaging. Most of these cases were diagnosed with BPS on MRI imaging. However, these results are not sufficient for generalization. The experience of the clinician performing the US imaging or the radiologist evaluating the MRI scans may affect prenatal diagnoses.

Study Limitations

This study had some limitations. The diagnosis of our cases could not be confirmed pathologically, since few of our cases were operated and most of the operated cases were operated in different centers. Additionally, CT was not preferred by clinicians in postnatal imaging of many cases. This study also has strength. This study included all outcomes of CLMs in the antenatal, perinatal, and postnatal periods. Additionally, it is one of the few studies that evaluated the association between US and MRI in prenatal imaging of cases.

CONCLUSION

Our study showed that CLMs are generally not associated with major structural and genetic anomalies. Although the cases showed mostly favorable prognosis in the antenatal period, adverse outcomes such as RDS and NICU admission were high in the perinatal period. Surgical resection was required in approximately 1/3 of the cases in the postnatal period.

Ethics

Ethics Committee Approval: The study protocol was approved by the University of Health Sciences Turkey, İzmir Tepecik Training and Research Hospital Local Institutional Ethics Committee (approval number: 2022/03-09, date: 15.03.2022).

Informed Consent: Retrospective study.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Concept: H.G., Design: H.G., Data Collection or Processing: C.G., İ.Ö., K.O.A., Analysis or Interpretation: C.G., İ.Ö., H.G.P., A.E., Literature Search: H.G., B.B., Writing: H.G., B.B.

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REFERENCES

1. Watarai F, Takahashi M, Hosoya T, Murata K. Congenital lung abnormalities: a pictorial review of imaging findings. Jpn J Radiol. 2012;30:787-97.
2. Berrocal T, Madrid C, Novo S, Gutiérrez J, Arjonilla A, Gómez-León N. Congenital anomalies of the tracheobronchial tree, lung, and mediastinum: embryology, radiology, and pathology. Radiographics. 2004;24:e17.
3. Newman B. Congenital bronchopulmonary foregut malformations: concepts and controversies. Pediatr Radiol. 2006;36:773-91.
4. Langston C. New concepts in the pathology of congenital lung malformations. Semin Pediatr Surg. 2003;12:17-37.
5. Biyyam DR, Chapman T, Ferguson MR, Deutsch G, Dighe MK. Congenital lung abnormalities: embryologic features, prenatal diagnosis, and postnatal radiologic-pathologic correlation. Radiographics. 2010;30:1721-38.
6. Stocker LJ, Wellesley DG, Stanton MP, Parasuraman R, Howe DT. The increasing incidence of foetal echogenic congenital lung malformations: an observational study. Prenat Diagn. 2015;35:148-53.
7. Adzick NS. Management of fetal lung lesions. Clin Perinatol. 2009;36:363-76.
8. Davenport M, Eber E. Long term respiratory outcomes of congenital thoracic malformations. Semin Fetal Neonatal Med. 2012;17:99-104.
9. Winters WD, Effmann EL. Congenital masses of the lung: prenatal and postnatal imaging evaluation. J Thorac Imaging. 2001;16:196-206.
10. Baird R, Puligandla PS, Laberge JM. Congenital lung malformations: informing best practice. Semin Pediatr Surg. 2014;23:270-7.
11. Calvert JK, Lakhoo K. Antenatally suspected congenital cystic adenomatoid malformation of the lung: postnatal investigation and timing of surgery. J Pediatr Surg. 2007;42:411-4.
12. Annunziata F, Bush A, Borgia F, et al. Congenital Lung Malformations: Unresolved Issues and Unanswered Questions. Front Pediatr. 2019;7:239.
13. Stocker JT, Madewell JE, Drake RM. Congenital cystic adenomatoid malformation of the lung. Classification and morphologic spectrum. Hum Pathol. 1977;8:155-71.
14. Daltro P, Werner H, Gasparetto TD, et al. Congenital chest malformations: a multimodality approach with emphasis on fetal MR imaging. Radiographics. 2010;30:385-95.
15. Mong A, Johnson AM, Kramer SS, et al. Congenital high airway obstruction syndrome: MR/US findings, effect on management, and outcome. Pediatr Radiol. 2008;38:1171-9.
16. Kunisaki SM, Fauza DO, Nemes LP, Barnewolt CE, Estroff JA, Kozakewich HP, Jennings RW. Bronchial atresia: the hidden pathology within a spectrum of prenatally diagnosed lung masses. J Pediatr Surg. 2006;41:61-5.
17. Barnes NA, Pilling DW. Bronchopulmonary foregut malformations: embryology, radiology and quandary. Eur Radiol. 2003;13:2659-73.
18. Gajewska-Knapik K, Impey L. Congenital lung lesions: Prenatal diagnosis and intervention. Semin Pediatr Surg. 2015;24:156-9.
19. Cavoretto P, Molina F, Poggi S, Davenport M, Nicolaides KH. Prenatal diagnosis and outcome of echogenic fetal lung lesions. Ultrasound Obstet Gynecol. 2008;32:769-83.
20. Crombleholme TM, Coleman B, Hedrick H, et al. Cystic adenomatoid malformation volume ratio predicts outcome in prenatally diagnosed cystic adenomatoid malformation of the lung. J Pediatr Surg. 2002;37:331-8.
21. Stanton M, Njere I, Ade-Ajayi N, Patel S, Davenport M. Systematic review and meta-analysis of the postnatal management of

congenital cystic lung lesions. J Pediatr Surg. 2009;44:1027-33.

22. Cobo T, Kacerovsky M, Jacobsson B. Risk factors for spontaneous preterm delivery. Int J Gynaecol Obstet. 2020;150:17-23.
23. Lee PA, Houk CP, Ahmed SF, Hughes IA; International Consensus Conference on Intersex organized by the Lawson Wilkins Pediatric Endocrine Society and the European Society for Paediatric Endocrinology. Consensus statement on management of intersex disorders. International Consensus Conference on Intersex. Pediatrics. 2006;118:e488-500.
24. Laberge JM, Puligandla P, Flageole H. Asymptomatic congenital lung malformations. Semin Pediatr Surg. 2005;14:16-33.
25. Aziz D, Langer JC, Tuuha SE, Ryan G, Ein SH, Kim PC. Perinatally diagnosed asymptomatic congenital cystic adenomatoid malformation: to resect or not? J Pediatr Surg. 2004;39:329-34.
26. Naito Y, Beres A, Lapidus-Krol E, Ratjen F, Langer JC. Does earlier lobectomy result in better long-term pulmonary function in children with congenital lung anomalies? A prospective study. J Pediatr Surg. 2012;47:852-6.
27. Papagiannopoulos KA, Sheppard M, Bush AP, Goldstraw P. Pleuropulmonary blastoma: is prophylactic resection of congenital lung cysts effective? Ann Thorac Surg. 2001;72:604-5.
28. Breysem L, Bosmans H, Dymarkowski S, et al. The value of fast MR imaging as an adjunct to ultrasound in prenatal diagnosis. Eur Radiol. 2003;13:1538-48.
29. Cannie M, Jani J, De Keyzer F, et al. Magnetic resonance imaging of the fetal lung: a pictorial essay. Eur Radiol. 2008;18:1364-74.