CASE REPORT / OLGU SUNUMU

DOI: 10.4274/atfm.galenos.2025.46704
J Ankara Univ Fac Med 2025;78(2):154-156

SURGICAL MEDICAL SCIENCES / CERRAHİ TIP BİLİMLERİ

Incidental Detection of Congenital Cystic Adenomatoid Malformation After Thoracoscopic Repair of Diaphragmatic Hernia

Torakoskopik Diyafram Hernisi Onarımı Sonrası Rastlantısal Konjenital Kistik Adenomatoid Malformasyonun Saptanması

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Abstract

Congenital pulmonary airway malformation (CPAM) is a rare benign lung lesion, and CPAM combined with congenital diaphragmatic hernia (CDH) is extremely rare. The patient being described was a neonatal boy with a left-sided CDH diagnosed after birth. Thoracoscopic repair was performed on the first postnatal day. On chest X-ray, a suspicious lesion in terms of recurrence was observed after the postoperative follow-up period. Type 2 CPAM was diagnosed based on computed tomography. The child is doing well on postoperative period and the lesion stays stable without any intervention. Congenital lung lesions with CDH are difficult to diagnose before the CDH repair. Although very rare, congenital lung lesions concurrent with CDH should be considered when managing these patients.

Keywords: Cystic adenomatoid malformation of lung, congenital, pulmonary surgical procedures, newborns, surgery, thoracoscopic

Öz

Konjenital pulmoner hava yolu malformasyonu (CPAM) nadir görülen benign bir akciğer lezyonudur ve konjenital diyafragma hernisi (KDH) ile birlikte CPAM oldukça nadirdir. Doğum sonrası sol taraflı konjenital diyafragma hernisi KDH tanısı alan yenidoğan erkek bir olgu sunulmuştur. Doğum sonrası birinci günde torakoskopik onarım yapıldı. Ameliyat sonrası takiplerinde çekilen akciğer grafisinde nüks açısından şüpheli lezyon görüldü. Tip 2 CPAM tanısı bilgisayarlı tomografi ile konuldu. Ameliyat sonrası dönemde çocuğun genel durumu iyi olup, lezyon herhangi bir müdahaleye gerek kalmadan stabil takip ediliyor. KDH ile birliktelik gösteren konjenital akciğer lezyonlarının KDH onarımından önce teşhis edilmesi zordur. Bu hastaların tedavisinde KDH ile birlikte nadir görülen konjenital akciğer lezyonları da göz önünde bulundurulmalıdır.

Anahtar Kelimeler: Akciğerin kistik adenomatoid malformasyonu, konjenital, akciğer cerrahisi işlemleri, yenidoğanlar, ameliyat, torakoskopik

Introduction

There are several theories explaining the development of pulmonary hypoplasia. Most accepted theory is that pulmonary hypoplasia occurs due to the mass effect of abdominal contents present in the thorax during lung development (1).

Structural defects with respiratory abnormalities which include congenital lung lesions such as congenital pulmonary airway malformation (CPAM) and bronchopulmonary sequestration (BPS) are rarely seen concurrent with congenital diaphragmatic hernia (CDH) (2). The presentation of an additional airway malformation rises concerns of morbidity with varying degrees of respiratory distress. There are also

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Received/Geliş Tarihi: 13.01.2024 **Accepted/Kabul Tarihi:** 06.01.2025 **Epub:** 02.05.2025 **Publication Date/Yayınlanma Tarihi:** 27.06.2025
Cite this article as/Atıf: İnal D, Bahadır K, Khalilova P, et al. Incidental detection of congenital cystic adenomatoid malformation after thoracoscopic repair of diaphragmatic hernia. J Ankara Univ Fac Med. 2025;78(2):154-156

studies demonstrating no difference in the outcome of CDH patients with concurrent congenital lung lesions (3). However, there is no specific data about the prognosis of CDH and CPAM.

The aim of this study is to present a child who has CDH with concurrent CPAM.

Case Presentation

Informed consent was obtained from the parents. A male newborn was born through cesarean section at the 39th week to a 27-year-old mother who was prenatally followed by an obstetrics and gynecology specialist. He had an apgar score of 6/8 at birth and developed respiratory distress in delivery room. The newborn was admitted to neonatal intensive care unit and nasal intermittent positive pressure ventilation was initiated. Arterial blood gas was evaluated to show normal results (power of hydrogen pH:: 7.34, pressure of carbon dioxide pCO_2: 39.8, lactate: 1.2). A chest X-ray was taken due to respiratory distress (Figure 1). Even though CDH was not prenatally diagnosed, signs of CDH were presented on the X-ray and the patient was rapidly intubated. Thorax ultrasound (US) imaging showed that the intestines and the colon was partially located in the left hemithorax. Echocardiography of the newborn revealed patent ductus arteriosus and pulmonary hypertension. Abdominal and urinary US of the newborn revealed no other anomalies. On the first postnatal day, thoracoscopic diaphragmatic hernia repair was performed. After the operation the newborn was stayed on mechanical ventilation and extubated on 3rd day of operation. The baby did not require any oxygen or respiratory support afterwards. In the first postoperative day, chest X-ray revealed a suspicious lesion (there is a multiloculated cystic lesion which was thought to be recurrent diaphragmatic hernia) in terms of recurrence was observed while the patient was clinically asymptomatic without any need for oxygen support (Figure 2). Distal colon imaging was performed which showed no recurrence. Thorax computed tomography (CT) was performed, and a suspicious cystic lesion was seen to be compatible with type 2 CPAM (Figure 3). The patient was referred to pediatric pulmonologist for follow-up.

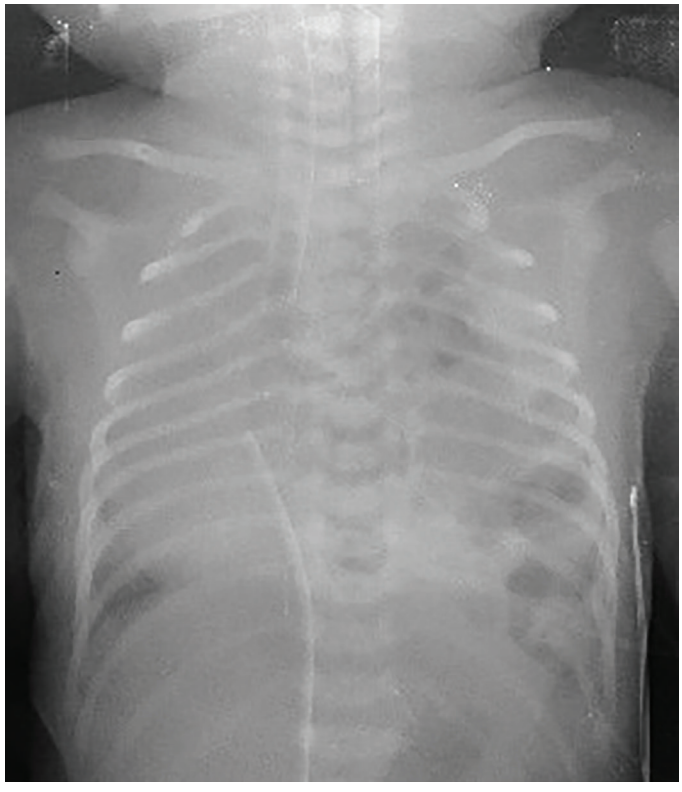

Figure 1: Chest X-ray which demonstrates congenital diaphragmatic hernia on the left side. Right hemithorax looks radiopaque possibly due to mediastinal shift

Discussion

We report a rare case of CPAM concurrent with CDH which is diagnosed after thoracoscopic CDH repair. Postoperative chest X-ray showed a lesion suspicious of bowel on the left hemithorax, which supported recurrence of CDH. CT was performed to reveal there was no recurrence but type 2 CPAM in the left lower lobe of the newborn was presented.

CDH is a life threating pathology in newborns and a cause of death due to pulmonary hypoplasia and pulmonary hypertension (4). On the other hand, CPAM has lower mortality but it is a potential cause of infection and malignancy (5). CDH is a major congenital anomaly caused by failure of closure of the pleuroperitoneal cavity by the fusion of septum transversum and pleuroperitoneal folds (2). In cases of pulmonary sequestration concurrence, it is hypothesized that the formation of sequestration during the embryonic period

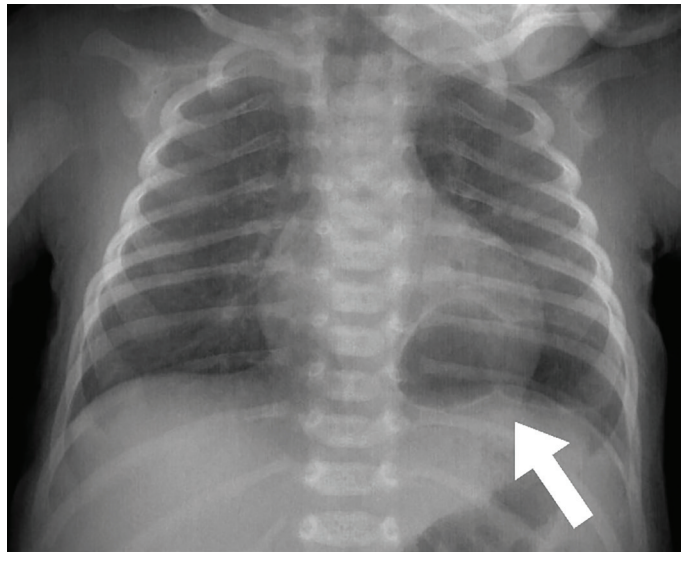

Figure 2: Chest X-ray shows suspicious lesion (there is a multiloculated cystic lesion which was thought to be recurrent diaphragmatic hernia) in the left lower zone

may interfere mechanically with the fusion of the lungs and the diaphragm (3).

In the literature, congenital lung lesions associated with CDH are rarely described and the exact incidence of these lesions concurrent with CDH is uncertain (2). Case series showed that the incidence of BPS with CDH are between 15 and 30% (4). Savic et al. (6) showed that the incidence of CDH with BPS were 3% and 27%. On the other hand, Soni et al. (4) showed that incidence of both CPAM and BPS with CDH was 7.2%. However, CPAM concurrent with CDH are reported very rarely.

CPAM is characterized by hamartomatous lesions of the lungs classified by the size of the cysts (7). Stocker's classification groups CPAM into three groups according to cyst size. Type 1 is the most common and has macrocystis larger than 2 cm. Type 2 which is associated with other congenital anomalies, has multiple cysts smaller than 2 cm. Type 3 has cysts smaller than 0.5 cm and includes solid components (7). In this case, type 2 CPAM was reported on CT following the operation for CDH. When CPAM appears as a single, localized, solid or cyst like lesion, it may be difficult to differentiate it from bronchogenic cysts, bronchial atresia and pulmonary sequestration (8).

The incidence rate of 5-65% for recurrence after CDH repair is reported in literature (9). The postoperative chest X-ray was suspicious of recurrence thus, a CT was performed to confirm diagnosis. CT showed two pulmonary cysts less than 2 cm in the left lower lobe, which was concordant with the lesion on the chest X-ray (Figure 3).

Surgery is a preferred method of treatment for symptomatic CPAM, but postnatal management of asymptomatic lesions are controversial (10). In the literature, some authors argue that early and elective thoracoscopic surgery prevent infective complications (11). By contrast, some authors argue that surgery is chosen based on patient according to the onset of symptoms (10).

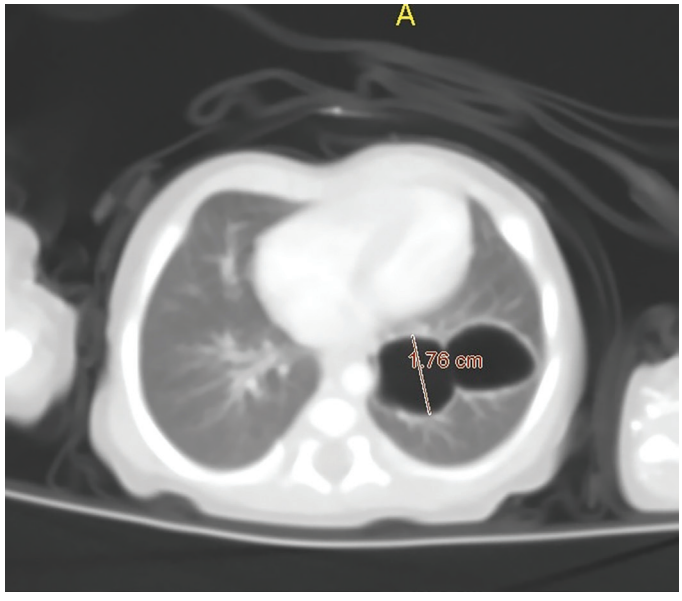

Figure 3: Axial CT scan of chest demonstrates a left lower lobe lesion with two cysts

CT: computed tomography

Congenital lung lesions with CDH are difficult to diagnose before the CDH repair. However, observations on the development of the lungs can be made by surgeons during the CDH repair. Although very rare, congenital lung lesions concurrent with CDH should be considered when managing these patients.

Ethics

Informed Consent: Informed consent was obtained from the parents.

Footnotes

Authorship Contributions

Surgical and Medical Practices: D.İ., P.K., E.E., U.A., Concept: D.İ., K.B., P.K., E.E., Design: D.İ., K.B., E.E., Data Collection or Processing: D.İ., E.E., U.A., Literature Search: D.İ., K.B., E.E., U.A., Writing: D.İ., K.B., P.K., E.E.

Conflict of Interest: The authors declare no conflict of interest.

Financial Disclosure: No financial assistance was received to support this study.

References

1. Dingeldein M. Congenital diaphragmatic hernia: management & outcomes. Adv Pediatr. 2018;65:241-247.
2. Kosiński P, Wielgoś M. Congenital diaphragmatic hernia: pathogenesis, prenatal diagnosis and management-literature review. Ginekol Pol. 2017;88:24-30.
3. Grethel EJ, Farrell J, Ball RH, et al. Does congenital diaphragmatic hernia associated with bronchopulmonary sequestration portend a better prognosis? Fetal Diagn Ther. 2008;23:250-253.
4. Soni S, Moldenhauer JS, Rintoul N, et al. Perinatal outcomes in fetuses prenatally diagnosed with congenital diaphragmatic hernia and concomitant lung lesions: A 10-year review. Fetal Diagn Ther. 2020;47:630-635.
5. Wagner R, Li H, Ayoub L, et al. Epithelial cell-adhesion protein cadherin 26 is dysregulated in congenital diaphragmatic hernia and congenital pulmonary airway malformation. Pediatr Surg Int. 2021;37:49-57.
6. Savic B, Birtel FJ, Tholen W, et al. Lung sequestration: report of seven cases and review of 540 published cases. Thorax. 1979;34:96-101.
7. Sintim-Damoa A, Cohen HL. Fetal imaging of congenital lung lesions with postnatal correlation. Pediatr Radiol. 2022;52:1921-1934.
8. Aljarad B, Alkhayer I, Alturk A, et al. A rare case of congenital pulmonary airway malformation in a 14-year-old male presenting with spontaneous pneumothorax. Ann Med Surg (Lond). 2021;68:102692.
9. Macchini F, Raffaeli G, Amodeo I, et al. Recurrence of congenital diaphragmatic hernia: risk factors, management, and future perspectives. Front Pediatr. 2022;10:823180.
10. Leblanc C, Baron M, Desselas E, et al. Congenital pulmonary airway malformations: state-of-the-art review for pediatrician's use. Eur J Pediatr. 2017;176:1559-1571.
11. Parikh DH, Rasiah SV. Congenital lung lesions: postnatal management and outcome. Semin Pediatr Surg. 2015;24:160-167.

CASE REPORT / OLGU SUNUMU

DOI: 10.4274/atfm.galenos.2025.85579
J Ankara Univ Fac Med 2025;78(2):157-160

MEDICAL SCIENCES / DAHİLİ TIP BİLİMLERİ

SARS-CoV-2 and *Mycobacterium Fortuitum* Coinfection: A Case Report

SARS-CoV-2 ve *Mycobacterium Fortuitum* Koenfeksiyonu: Olgu Sunumu

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Abstract

Coronavirus disease-2019 (COVID-19) pandemic caused millions of people to become infected and had resulted several deaths. After initial resolution, in cases of clinical deterioration, it is essential to consider the possibility of coinfections. Non-tuberculous mycobacteria infections are rare and often overlooked. In this report, we present a severe acute respiratory syndrome-Coronavirus-2 and *Mycobacterium fortuitum* coinfected patient. Our intention is to bring attention to the possibility of such coinfection without a known history of any lung diseases or immunosuppression other than COVID-19 and thus, broadening the clinical thinking process of physicians.

Keywords: SARS-CoV-2, non-tuberculous mycobacteria, COVID-19

Öz

Koronavirüs hastalığı-2019 (COVID-19) pandemisi milyonlarca insanın enfekte olmasına ve çok sayıda ölüme neden olmuştur. Başlangıçtaki iyileşme döneminin ardından klinik kötüleşme olması halinde koenfeksiyon olasılığını düşünmek gerekmektedir. *Tüberküloz dışı mikobakteri* enfeksiyonları nadir olup sıklıkla gözden kaçırılmaktadır. Bu yazıda şiddetli akut solunum sendromu-Koronavirüs-2 ve *Mycobacterium fortuitum* koenfeksiyonu olan bir hasta sunmaktayız. Amacımız, eşlik eden akciğer hastalığı ve COVID-19 dışında immünosüpresyon öyküsü bulunmadığında da bu koenfeksiyonun gelişebileceğine dikkat çekmek ve hekimlerin klinik yaklaşımına katkı sağlamaktır.

Anahtar Kelimeler: SARS-CoV-2, *tüberküloz dışı mikobakteri*, COVID-19

Introduction

There are nearly 200 species of non-tuberculous mycobacteria (NTM), most of which live in soil and water in rural and urban areas (1). Almost half have been associated with opportunistic infections in animals and humans, causing sporadic outbreaks (1). NTM is acquired through exposure to water, aerosols, soil and dust via inhalation, ingestion, cracks due to skin injuries, surgical procedures, or catheterization (1). Almost all patients with NTM pulmonary disease have chronic or recurring cough. Other symptoms include sputum, fatigue, malaise, dyspnea, fever, hemoptysis, chest pain, and weight loss (1). Both 2020 "Treatment of Non-tuberculous Mycobacterial Pulmonary Disease" clinical practice guideline and expert panel group for management recommendations in NTM pulmonary diseases recommend using clinical (pulmonary symptoms, and

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Received/Geliş Tarihi: 12.02.2025 **Accepted/Kabul Tarihi:** 19.04.2025 **Epub:** 30.05.2025 **Publication Date/Yayınlanma Tarihi:** 27.06.2025

Cite this article as/Atıf: Gül T, Maraş H, Demir Ö, et al. SARS-CoV-2 and *mycobacterium fortuitum* coinfection: a case report. J Ankara Univ Fac Med. 2025;78(2):157-160

***This case report was presented as an oral presentation during the Turkish Society of Clinical Microbiology and Infectious Diseases COVID-19 Symposium (10-12 September, 2021, İstanbul).**