Jurnal 39 Scopus Q2 Left Pulmonary Aplasia A Case Report

by Turnitin.com ®

Submission date: 08-Apr-2022 02:52PM (UTC+0700)
Submission ID: 1805096970
File name: Jurnal_39_Scopus_Q2_Left_Pulmonary_Aplasia_A_Case_Report.pdf (836.82K)
Word count: 2444
Character count: 14239

ISSN 0975-2366
DOI: https://doi.org/10.31838/ijpr/2020.12.03.307

Research Article

Left Pulmonary Aplasia: A Case Report

HERLINA UINARNI[1,2*], MARIA ANGELINA YASHINTA G[2], LILIANA SUGIHARTO[1], HULDANI[3], HARUN ACHMAD[4], ALEXANDER MARKOV[5], DMITRY O. BOKOV[6]

[1]Department of Anatomy, School of Medicine and Health Sciences Atma Jaya Catholic University of Indonesia.
[2]Department of Radiology Grand Family Mother and Child Care Hospital, Indonesia
[3]Department of Physiology, Faculty of Medicine, Lambung Mangkurat University, Banjarmasin, South Kalimantan, Indonesia.
[4]Department of Pediatric Dentistry, Faculty of Dentistry, Hasanuddin University, Makassar, South Sulawesi, Indonesia.
[5]Tyumen State Medical University, Tyumen, Russian Federation
[6]Sechenov First Moscow State Medical University, Moscow, Russian Federation
*Corresponding Author
Email ID: herlina.uinarni@atmajaya.ac.id.
Received: 11.03.20, Revised: 12.04.20, Accepted: 13.05.20

ABSTRACT
We reported a death case of a baby girl due to symptomatic left pulmonary aplasia. Pulmonary aplasia or agenesis is a rare case of pulmonary underdevelopment. Multimodality imaging have been used for diagnosis and follow up of the bronchopulmonary malformation from antenatal to postnatal period. No treatments needed in asymptomatic cases. Oxygen therapy, rehabilitation, prevention and treatment of infections, bronchodilators and other treatments are required in symptomatic cases. Surgical procedure may be done to stump removal if postural drainage and antibiotics fail to resolve the infection. Prognosis depends on many factors, includes the severity of the congenital anomalies, involvement and functionality of the normal lung in any disease process, and many others.

Keywords: pulmonary aplasia, pulmonary agenesis, congenital pulmonary malformation, neonatal chest ultrasound.

BACKGROUND

Pulmonary underdevelopment is classified into 3 entities : pulmonary aplasia, agenesis and hypoplasia.[1,2] The failure of bronchial analogue to divide equally between two lungs with possible abnormal blood flow in dorsal aortic arch during this period may result in hypoplasia, aplasia and agenesis of unilateral pulmonary parenchyma.[3] Pulmonary agenesis and aplasia are rare abnormality with incidence between 0.0034% and 0.0097%[4,5]. We reported a case of pulmonary aplasia with early onset neonatal symptoms.

CASE PRESENTATION

A female baby was born by caesarean delivery at 35 weeks of gestation to a 26-year-old mother, with low birth weight 1890 gram. The pregnancy was uneventful and there was no relevant family history. The Apgar scores were 7 and 8 at 1 and 5 min, respectively. At 8 hours of life, she became tachypneic, chest wall movement was asymmetric. Chest radiography revealed complete left lung field opacity, mediastinal shift towards affected side, left hemithorax volume loss and crowding of ribs. Chest ultrasound was performed and showing no pleural sliding sign on the left side. Left hemithorax was filled with heart and fluid. Left diaphragm was elevated. Immunoserology examination was positive for toxoplasma and rubella IgG. Another significant finding is buphthalmus and congenital glaucoma on the left eye. Despite the therapy given, the infant showed deterioration, and died at 8 days old.

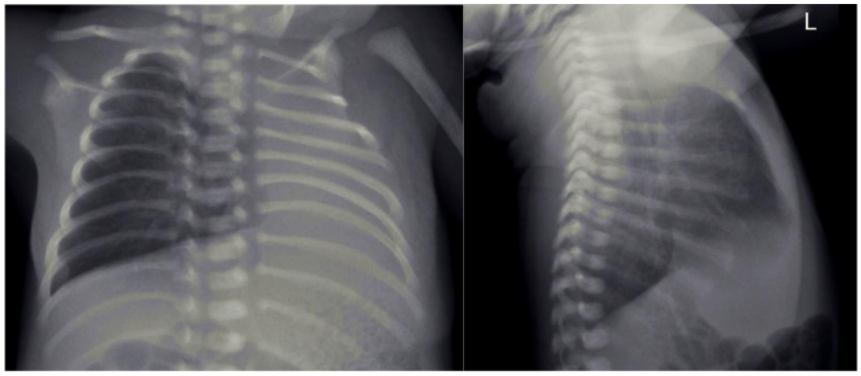

Fig.1: Chest x-ray at 8 hours old neonate showingopacification of left hemithorax, mediastinal shift to affected side, crowding of left ribs. Right lung was hyperinflated, mildly herniated to the left side. Left diaphragm contour was unclear. Increased retrosternal lucency is depicted on lateral view.

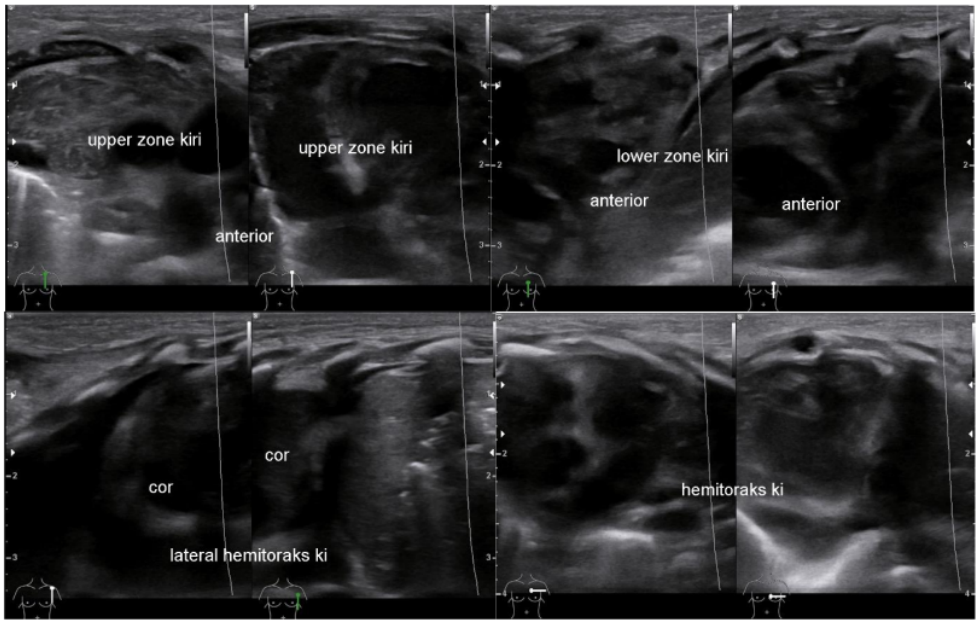

Fig.2: Chest ultrasound showing left hemithorax was filled with heart and other mediastinal structure. Pleural fluid is noted around the mediastinal structure.

DISCUSSION

The intrauterine development of the human lunghas been divided into five phases: embryonic, pseudoglandular, canalicular, saccular, and alveolar. The embryonic phase begins during the 4th week of gestation with the formation of the respiratory diverticulum (laryngotracheal bud) from the ventral wall of the primitive foregut.[2,3,4]

For the first time pulmonary agenesis was classified by Schneider which later on was modified by Boyden into three groups according to development of their primitive lung bud. Type I which is called pulmonary agenesis is complete absence of unilateral lung parenchyma, its bronchus and vasculature. Type II is named pulmonary aplasia which is complete absence of unilateral lung with a rudimentary bronchus. Type III is pulmonary hypoplasia characterized by hypoplastic bronchial tree and pulmonary artery with a variable amount of lungparenchyma.[1,3] In

both lung agenesis and aplasia, the ipsilateral pulmonary artery is absent.[5,6]

The main etiology of the disease is unknown. Genetic, teratogenic, lack of vitamin A during pregnancy, viral agents, and mechanical factors may have a bearing on etiology.[3,4] It has been hypothesized that abnormal blood flow in the dorsalaortic arch during the 4th week of gestation (embryonic phase) causes pulmonary agenesis.[2,3]

The clinical features vary from asymptomatic to variable respiratory complaints such as dyspnea and respiratory distress with history of recurrent chest infections. According to the literature, left side agenesis is more common comparing to the right side with longer life expectancy.The symptoms may occur as early as in neonatal period or later on during childhood and even adult life.[3] The majority of patients initiate with symptoms during the first year oflife.[5] Reasons attributed to signs of respiratory insufficiency in these babies may be due to kinking and compression of trachea, mediastinal shift and posterior curvature of trachea due to pressure by dislocation of aortic arch and truncusarteriosus. Spillage from rudimentary bronchus may attribute to reccurent infection.[6,7]

Physical examination of patients shows asymmetric chest wall movements with absent or decrease respiratory sounds in unilateral hemithorax.[3] More than50% of affected fetuses have other abnormalities involving the cardiovascular (patent ductusarteriosus, patent foramen ovale), gastrointestinal(tracheoesophageal fistula, imperforate anus),genitourinary, or skeletal (limb anomalies, vertebral segmentation anomalies) system, facial or ear abnormalities.[2,6]

Diagnosis and follow up of the bronchopulmonary malformations are performed with
different imaging techniques (ultrasound, plain radiograph, MR, CT).[8] Congenital pulmonary malformations are increasingly being detected with prenatal ultrasonography (US), often supplemented by magnetic resonance (MR) imaging.[9] Prenatal Ultrasound is an excellent initial screening test and the Doppler mode allows to know the lesion vasculature.[8] At antenatal ultrasound, fetal lungs appear as homogeneous and are slightly more echogenic than the liver. Theechogenicity of the lung increases as gestationadvances. Fetallung volumes can be measured with three and four dimensional US and should be calculated infetuses with lung abnormalities for the estimationof residual lung volume. Unilateral pulmonary agenesis is diffcult to diagnosewith prenatal US; however, it can be suspectedon the basis of mediastinal shift.[2]

Plain radiograph is the initial imaging for diagnosis of pulmonary underdevelopment using 2-view chest radiograph.[7,8] Imagingfindings in pulmonary aplasia and agenesis aresimilar, except for the presence of a short blindending bronchus in aplasia. Postnatal radiographydemonstrates small, diffuse opacification of the involvedhemithorax with ipsilateralmediastinal shift, and elevated hemidiapraghm. The normal contralateral lung shows compensatory hyperinflation and herniation across the anterior midline, which is best seen on the lateral projections as a band of increased retrosternal lucency.[1,2,5]

US is a practical imaging modality, due to its widespread availability, relative ease of performance, and lack of ionizing radiation exposure. It is useful for assessing focal congenital pulmonary malformations such as foregut duplication cysts or pulmonary sequestration in neonates and young infants.[9] Its usefull in pulmonary underdevelopment had not been reported. In this patient, we use ultrasound to look for sign of normal aerated lung, such as pleural sliding, A line and B line on the affected hemithorax. To achieve optimal sonographic imaging evaluation in neonates and young infants, a high-resolution 10 to 15-MHz linear-array transducer in a transsternal, parasternal, or intercostal approach can be used. In characterizing the lesions, imaging in at least 2 planes is recommended, and color flow may aid in demonstrating associated anomalous vessels.[9]

Multidetector CT with multiplanar 2D and 3D imaging helps confirm the absence of the lung parenchyma, bronchus, and pulmonary artery on the involved side by clearly identifying the bronchial stump and/or the rudimentary bronchial tree.[1,2] CT also depict the degree of mediastinal displacement, size of the contralateral pulmonary artery, vascular compression of the trachea, and ipsilateralpleural fluid collection.[6,10]

No treatment is required in asymptomatic cases. Treatment consists of supportive measures such as oxygen therapy, rehabilitation (to improve clearing of secretions), prevention and treatment of infections, bronchodilators and controlling other complications. Patients having stumps may require surgical removal of the stump if postural drainage and antibiotics fail to resolve the infection. Corrective surgery of associated congenital anomalies, wherever feasible, may be undertaken.[3,4,5,11]

Prognosis depends on severity of associated congenital anomalies, involvement and functionality of the normal lung in any disease process, infection of the single lung, pulmonary arterial hypertension, and kinking of the large

vessels due to mediastinal displacement. Pulmonary hypertension in these patients is due to normal blood volume flow through a reduced pulmonary vascular bed.[12,13,14] Patients with right lung agenesis have a higher mortality than those with left lung agenesis because of compression of the tracheobronchial tree by the shifting of normally midthoracic structures into the right chestand greater rotation of the trachea, heart and large vessels.[3,4,5,7,15,16] If patient survives the first five years without major infection, an almost normal life span can be expected.[3,4,5,17,18,19,20]

REFERENCES

1. Epelman M, Daltro P, Soto G, Ferrari CM, and Lee EY. Congenital Lungs Anomalies. In : Coley BD editors. Caffey's Pediatric Diagnostic Imaging. Twelfth Edition. Philadelphia: Saunders; 2013.p.550
2. 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(6):1721–38
3. Sadiqi J, Hamidi H. CT features of lung agenesis – a case series (6 cases). BMC Medical Imaging. 2018;18:37
4. Shrestha P, Poudel P, Shah PL. Unilateral pulmonary aplasia: a case report. J Nepal Paediatr Soc. 2010;30:116-118
5. Olivares AIS, Grub JP, Balderas LJ, González CJ, Gómez JS, Bechara JK. Pulmonary aplasia: two case reports. Bol Med Hosp Infant Mex. 2015;72(1):66-70
6. Patterson A. Imaging Evaluation of Congenital Lung Abnormalities in Infants and Children. RadiolClin N Am 2005; 43:303 – 323
7. Khurram MSA, Rao SP, Vamshipriya A. Pulmonary agenesis: A case report with review of literature. Qatar Medical Journal. 2013; 14:38-40
8. Traba OS, Vigara AP, Pajares MP, Zurita MB, Larrucea JAT, Arellano JP. Congenital Lung Malformations, what the radiologist should know. ECR. 2013;C-0457
9. Lee EY, Dorkin H, Vargas SO. Congenital pulmonary malformations in pediatric patients: review and update on etiology, classification, and imaging findings. RadiolClin North Am. 2011;49:921-948.
10. Herlina Uinarni, Conny Tanjung, Huldani, Aminuddin Prahatama Putra, Mashuri, Bayu Indra Sukmana, Heru Wahyudi, Abdullah Zuhair, Aderiel Gabrian Tarius, Wisnu Wiryawan, Renie Kumala Dewi, Yunita Feby Ramadhany, Harun Achmad. The Importance of Ultrasound Findings in Children with Acute Abdominal Pain to Prevent Unnecessary Surgery. SRP. 2020; 11(4): 377-383. doi:10.31838/srp.2020.4.56
11. Huldani, Ilhamjaya Pattelongi, Muhammad Nasrum Massi, Irfan Idris, Agussalim Bukhari, Agung Dwi Wahyu Widodo, Herlina Uinarni, Austin Bertilova Carmelita, Adelgrit Trisia,San Gunma, Bagas Kara Adji Prayudhistya, Harun Achmad. Cortisol, IL-6, TNF Alfa, Leukocytes and DAMP on Exercise. SRP. 2020; 11(6): 474-485. doi:10.31838/srp.2020.6.74
12. Huldani, Mohammad Rudiansyah, Fauzie Rahman, Adelgrit Trisia, Sri Ramadhany, Siti Kaidah, Harun Achmad, Bayu Indra Sukmana, Dhea M Swengly, Shelsy Marippi, Wafa Ahdiya, Muhammad Hasan Ridhoni, Amalia Rahman, Zhasifa Khoirunnisa Suwanto, Gandhi Mahesa Priambodo, Muhammad Rafagih, Abdullah Zuhair. The Influence of Uric Acid Levels on Blood Pressure and Chronic Hypertension towards Hypertension Patient Proteinuria Levels (Overview of the Banjar Ethnic at the Cempaka Banjarmasin Health Center). SRP. 2020; 11(5): 52-56. doi:10.31838/srp.2020.5.09
13. Huldani, Siti Kaidah, Dwi Laksono Adiputro, Harun Achmad, Bayu Indra Sukmana, Deby Kania Tri Putri, Yusrinie Wasiaturrahmah, Renie Kumala Dewi, Didit Aspriyanto, Isnur Hatta, Saka Winias, Ariyati Retno Pratiwi, Ernita sari, Aminuddin Prahatama Putra, Anastasya Deborah M. C Manik, Ketrin Zailin, Ika Kusuma Wardani. Effect of Total Cholesterol Levels and Triglycerides on Blood Pressure Hypertension Patients Overview against Puskesmas Banjar Ethnic Group in Cempaka Banjarmasin. SRP. 2020; 11(4): 384-389. doi:10.31838/srp.2020.4.57
14. Mohammad Rudiansyah, Leonardo Lubis, Ria Bandiara, Bernadet Maria Sanjaya, Hendra Wana Nur'amin, Huldani, Abdul Hadi Martakusumah, Harun Achmad, Rully Marsis Amirullah Roesli, Dedi Rachmadi. The Correlation between Fibroblast Growth Factor 23 (FGF23) and Iron Profile in Chronic Kidney Disease Patients on Dialysis with Anemia. SRP. 2020; 11(6): 780-784. doi:10.31838/srp.2020.6.114
15. Harun Achmad, Andi Mardiana Adam, Aulia Azizah, Bayu Indra Sukmana, Huldani, Shafira Nurul Khera, Yunita Feby Ramadhany. A Review of Bandotan Leaf Extract (Ageratum conyzoides L.) in Inhibition Test to the Growth of Bacteria (Porphyromonas gingivalis) Case of Periodontitis Disease. SRP. 2020; 11(4): 390-395. doi:10.31838/srp.2020.4.58
16. Harun Achmad, Sri Oktawati, Andi Mardiana Adam, Burhanuddin Pasiga, Rizalinda Sjahril, Aulia Azizah, Bayu Indra Sukmana, Huldani, Heri Siswanto, Ingrid Neormansyah. Granulicatella Adiacens Bacteria Isolation from Perodontitical Patients with Polymerase Chain Reaction Techniques. SRP. 2020; 11(4): 394-400. doi:10.31838 /srp. 2020. 4.59
17. Huldani, Ilhamjaya Pattelongi, Muhammad Nasrum Massi, Irfan Idris, Agussalim Bukhari,

Agung Dwi Wahyu Widodo, Harun Achmad. Research Reviews on Effect of Exercise on DAMP's, HMGB1, Proinflammatory Cytokines and Leukocytes. SRP. 2020; 11(4): 306-312. doi:10.31838/srp. 2020. 4.44

18. Huldani, Harun Achmad, Aryadi Arsyad, Aminuddin Prahatama Putra, Bayu Indra Sukmana, Dwi Laksono Adiputro, Julia Kasab. Differences in VO2 Max Based on Age, Gender, Hemoglobin Levels, and Leukocyte Counts in Hajj Prospective Pilgrims in Hulu Sungai Tengah Regency, South Kalimantan. SRP. 2020; 11(4): 09-14. doi:10.31838/ srp.2020.4.03
19. Huldani, Sukmana BI, Pujiningtyas A, Savitri E, Fauziah, Nihayah U. (2019). Cellular Immunity of River Water Consumption and Bandarmasih Municipal Waterworks Consuments. Indian Journal of Public Health Research and Development. 10 (7): 789-94. DOI: http://dx.doi.org/10.5958/0976-5506.2019.01674.7
20. Huldani, Asnawati, Auliadina D, Amilia, FR, Nuarti N, Jayanti R. (2019). Abdominal Circumference, Body Fat Percent, and VO2 Max in Pilgrims of Hulu Sungai Tengah Regency. Journal of Physics: Conference Series. 1374 (1). https://doi.org/10.1088/1742-6596/1374/1/012058

Jurnal 39 Scopus Q2 Left Pulmonary Aplasia A Case Report

ORIGINALITY REPORT

15%	12%	8%	4%
SIMILARITY INDEX	INTERNET SOURCES	PUBLICATIONS	STUDENT PAPERS

PRIMARY SOURCES

1	Sujith V. Cherian, Anupam Kumar, Daniel Ocazionez, Rosa M. Estrada -Y- Martin, Carlos Santiago Restrepo. "Developmental lung anomalies in adults: A pictorial review", Respiratory Medicine, 2019 Publication	2%
2	Submitted to Universitas Hasanuddin Student Paper	2%
3	medical-case-reports.imedpub.com Internet Source	1%
4	docplayer.net Internet Source	1%
5	archive.org Internet Source	1%
6	journals.lww.com Internet Source	1%
7	www.hindawi.com Internet Source	1%

8	Trotman-Dickenson, Beatrice. "Congenital Lung Disease in the Adult : Guide to the Evaluation and Management", Journal of Thoracic Imaging, 2015. Publication	1 %
9	academic.oup.com Internet Source	1 %
10	bmcpregnancychildbirth.biomedcentral.com Internet Source	1 %
11	coek.info Internet Source	1 %
12	"Congenital Disorders of the Lung", Nelson Textbook of Pediatrics, 2011. Publication	1 %
13	ejmcm.com Internet Source	1 %
14	link.springer.com Internet Source	1 %
15	doaj.org Internet Source	1 %

Exclude quotes	Off	Exclude matches	Off
Exclude bibliography	Off		

Jurnal 39 Scopus Q2 Left Pulmonary Aplasia A Case Report

GRADEMARK REPORT

FINAL GRADE

/0

GENERAL COMMENTS

Instructor

PAGE 1

PAGE 2

PAGE 3

PAGE 4

PAGE 5