

Diaphragmatic Dysfunction after Open Heart Surgery on Infant: A Case Report

Dionisius Nico Oetomo¹, Fajar Perdhana² and Fitria Hasdiana³

¹Department of Anesthesiology and Reanimation, Faculty of Medicine – UNIVERSITAS AIRLANGGA, Surabaya, Indonesia

²Department of Anesthesiology and Reanimation, Dr Soetomo General Academic Hospital, Surabaya, Indonesia

³Faculty of Medicine – Universitas Airlangga, Surabaya, Indonesia

¹nico.oetomo123@gmail.com, ²dr.fajarperdhana@gmail.com and ³fitriahasdiana@gmail.com

Cite this paper as: Dionisius Nico Oetomo, Fajar Perdhana and Fitria Hasdiana (2024). Diaphragmatic Dysfunction after Open Heart Surgery on Infant: A Case Report. *Frontiers in Health Informatics*, 13(3), 5563-5569

ABSTRACT

This case study aims to understand the diaphragmatic dysfunction that occurs after open heart surgery, which often results from injury to the phrenic nerve. This dysfunction has a significant impact on the breathing capacity of patients, especially children undergoing such procedures. Methods of examining diaphragm dysfunction can be done by clinical observation, laboratory evaluation and fluoroscopic imaging analysis in pediatric patients with complex congenital heart defects. The results of this case study show that phrenic nerve injury causes diaphragmatic paralysis, thus slowing down the healing process and prolonging the need for mechanical ventilation in patients. Appropriate early management, including diaphragmatic plication intervention, can accelerate the ventilator weaning process and reduce the complications of pulmonary infection. These findings highlight the importance of close monitoring of patients' respiratory function postoperatively and provide insight into diaphragm injury management strategies to improve clinical outcomes.

Keywords: *Diaphragm Dysfunction, Phrenic Nerve, Open Heart Surgery*

INTRODUCTION

The diaphragm is a dome-like structure made of fibromuscular tissue that separates the thoracic cavity from the abdominal cavity and is an important muscle for breathing. Embryologically the diaphragm is formed by four components, namely the septum transversum, pleuroperitoneal folds, esophageal mesentery and body wall muscles. The formation of the diaphragm occurs in the 4th-12th week of pregnancy [1]. Normally these four components come together, but sometimes this fusion occurs incompletely.

Open Heart Surgery is one of the most common causes of PNI (Phrenicus Nerve Injury) in children, with an incidence of between 0.3% and 12.8%. Recent studies have shown the incidence of postoperative diaphragmatic paralysis due to PNI to be between 0.46% and 5.5% of patients [2].

Phrenic nerve dysfunction is a condition that often results from certain medical procedures, especially heart surgery. One of the main causes is diaphragm paralysis, which can occur in 2% to 20% of postoperative patients. This condition can be triggered by "phrenic frostbite," which is nerve damage due to the use of cold cardioplegia during coronary bypass surgery or stenting in heart valve surgery. In addition, phrenic nerve dysfunction can also be caused by direct invasion of the tumor into the nerve. Other factors that cause phrenic nerve neuropathy include infectious infections such as shingles and Lyme disease, immunological disorders such as Guillain-Barré syndrome, as well as metabolic conditions such as diabetes. Radiation therapy is also known to impair the function of the phrenic nerve, thus affecting the performance of the diaphragm [3].

The clinical manifestations between diaphragmatic eventration and diaphragmatic hernia are often difficult to distinguish, both clinically and radiologically. In many cases, diaphragmatic eventration is accompanied by other abnormalities, which can complicate the diagnosis. In this context, a correct diagnosis is essential to determine the appropriate course of treatment.

In diagnosing diaphragmatic eventration, the history of previous surgeries, especially those related to injury to the phrenic nerve, may provide important clues. Phrenic nerve injury often leads to diaphragmatic paralysis, which can be seen in cases of eventration. Furthermore, congenital eventration is more common in the left hemidiaphragm than the right, which can be identified through radiological examinations such as thorax X-ray. On X-ray, a diaphragm that appears higher with a smooth surface without any obvious defect is often indicative. Fluoroscopy is also used to differentiate eventration from diaphragmatic hernia, where eventration will show paradoxical movement of the diaphragm. Ultrasound is also used to look at the thickness of the diaphragm and evaluate other organ abnormalities, such as cardiac abnormalities [4]–[6].

Further assessment is done through phrenic nerve stimulation, which is the gold standard in diagnosing this nerve injury. However, fluoroscopy is considered more sensitive than phrenic nerve stimulation in detecting injury. This is important as the management of phrenic nerve injuries has two main approaches, namely conservative and invasive. In asymptomatic patients, rehabilitation therapy and physiotherapy are routinely used to monitor the condition. However, in symptomatic patients, ventilator support is required for 4-6 weeks while awaiting possible spontaneous resolution of the phrenic nerve injury [7].

Diaphragmatic plication is a surgical procedure often chosen to treat diaphragmatic eventration, especially in children under one year of age. It aims to flatten the abnormal diaphragm by placing sutures on the diaphragmatic dome. This approach can be done through the abdominal cavity or thoracic cavity, depending on the side of the diaphragm involved. The intrathoracic approach is more commonly used for the right diaphragm to avoid injury to important organs such as the liver, while for the left diaphragm, the approach is more at risk of injuring the abdominal cavity organs. If eventration occurs bilaterally, plication is performed via an intra-abdominal approach [8]. While supportive ventilation was previously the preferred treatment for phrenic nerve injury (PNI) after open heart surgery, diaphragmatic plication is now the most widely accepted therapy, especially in children under 1 year of age [9].

The main benefits of diaphragm plication include speeding up ventilator discharge, reducing the risk of pulmonary infection, and shortening the length of stay (LOS). Diaphragmatic plication is now the primary therapeutic option for patients with phrenic nerve injury post-open heart surgery, replacing supportive ventilation as the more widely accepted therapy, especially in children [10].

CASE STUDY

The patient, a one-year-old infant, was diagnosed with congenital heart disease at five months of age. Initial symptoms reported by the patient's mother included the patient's inability to feed well, frequent interruptions in feeding, and blue nail color. The patient also often appeared bluish, especially when crying for a long time or when feeling cold, but these symptoms gradually improved when the patient calmed down. Currently, the patient has a good appetite and drink, but has not gained weight for seven months of age. In addition, the patient is prone to coughs and colds.

Birth history showed that the patient was the second child, born by cesarean section at full-term gestation with a birth weight of 3,200 grams and a body length of 47 cm. After birth, the patient cried immediately, but there was a history of bluish body color without a history of jaundice. In terms of growth and development, the patient had difficulty gaining weight and was currently below the KMS curve. The patient's basic immunization history was complete, and no allergies were reported. The medication history noted the use of Propranolol at a dose of 3x4 mg as well as iron supplements at a dose of 1x0.7 ml per day.

The patient was in fair condition, with *compos mentis* consciousness. Pulse rate was measured at 124 beats per minute, regular, and respiratory frequency was 28 beats per minute. Oxygen saturation (SpO₂) was

recorded at 79%, with a body temperature of 36.8°C.

Clinical examination showed that the patient appeared cyanotic, but there was no anemia, jaundice, or shortness of breath. Thorax examination showed a symmetrical chest without retraction. Heart sounds S1 and S2 were single, regular, with a murmur but no gallop. On lung examination, vesicular breath sounds were heard on both sides, without rales or wheezing. The abdomen was soft with normal bowel sounds. Extremities were warm, dry, red in color with capillary filling time less than 2 seconds, and no edema.

RESULTS AND DISCUSSION

The results of the transthoracic echocardiogram (TTE) on October 2, 2023 showed some noteworthy findings. From the laboratory analysis, the hemoglobin (Hb) result of 16.4 g/dL and hematocrit (Ht) of 55.3% indicated a tendency of polycythemia, which may require further evaluation. The white blood cell (WBC) count was within normal limits, but the platelet count (PLT) was also within the normal range. Renal function parameters such as BUN (8.5 mg/dL) and creatinine (0.5 mg/dL) showed good renal function, while albumin (4.41 g/dL) and sodium, potassium and chloride (136/4.5/109) levels showed good electrolyte balance.

On TTE, there were situs inversus and dextrocardia with significant congenital variations, including AV concordance and VA discordance. Hypoplastic LV and unbalanced ventricle were also found. Although pulmonary venous drainage appeared normal, the aorta was located on the right. The aortic valve was normal without regurgitation, but there was severe infundibular pulmonary stenosis with a pressure gradient (PG) of 75 mmHg. In addition, there was a large primum ASD (1.64 cm) and a large inlet VSD (0.931 cm), indicating abnormal structural complexity of the heart. Left ventricular systolic function (EF) was measured at 77%, indicating reasonably good cardiac function despite the structural malformations. The overall conclusion includes a diagnosis of DORV (Double Outlet Right Ventricle) with large inlet VSD, single atrium, and other associated abnormalities, indicating the need for medical management and possibly surgical intervention in the future.

The conclusion of this examination showed that the patient had several complex congenital heart defects, namely Double Outlet Right Ventricle (DORV), Single Ventricle, Single Atrium, with Dextrocardia and Ambiguous Site characterized by Left Isomerism. The patient also had severe pulmonary stenosis in the infundibular and valvular parts, as well as Bilateral SVC and MAPCA. All of these results suggest a complex case of congenital heart defect that requires stepwise intervention to improve the patient's impaired blood circulation and oxygenation.

The treatment decision was to perform a PA banding procedure, non-pulsatile bilateral BCPS (Bilateral Cavopulmonary Shunt) with MPA ligation, and use CPB (Cardiopulmonary Bypass) on October 4, 2023. Postoperatively from October 4 to October 14, 2023, the patient was in the process of weaning the ventilator but spontaneous breathing was still inadequate. SPONT ventilator mode with PEEP 4, P_{support} 15, and FiO₂ 30%. achieved VT (tidal volume) 25-30, rate 35-40, MV (minute volume) 0.8-1.2, SpO₂ 88-90%. Vital signs showed ABP 62-74/38-45 mmHg fluctuating and heart rate 131 - 155 bpm. The low ABP with increasing heart rate indicates a hemodynamic condition that requires support, with additional nor-adrenaline and adrenaline pump to maintain perfusion and cardiac output, as we suspected LCOS (low cardiac output syndrome) or septic shock. Fluid accumulative balance was at a surplus of 25.67 cc and there was an increase in respiratory effort. Periodic chest Xray evaluation showed a significant change in left diaphragm height, suspected due to complications of PNI (Phrenicus Nerve Injury) post open heart surgery (Figure 1).

The patient was decided to change to PSIMV ventilator mode with parameters adjusted according to the patient's condition and periodic observation. Changes in the patient's condition such as decreased oxygen saturation (SpO₂) and increased airway pressure were monitored and regulated through changes in PEEP and FiO₂, including the administration of antibiotics (meropenem, ceftazidime), cardiac support drugs such as nor-adrenaline, adrenaline, and milrinone, and additional therapies such as nebulization and chest physiotherapy treatments to support lung function.

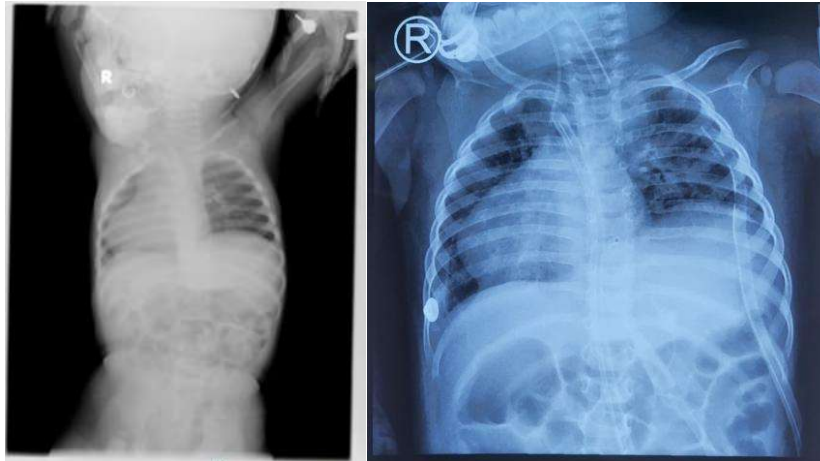


Figure 1: Pre-operation babygram, both diaphragms normal (Left), Chest x-ray post open-heart surgery, left diaphragm appears to be elevated (Right)

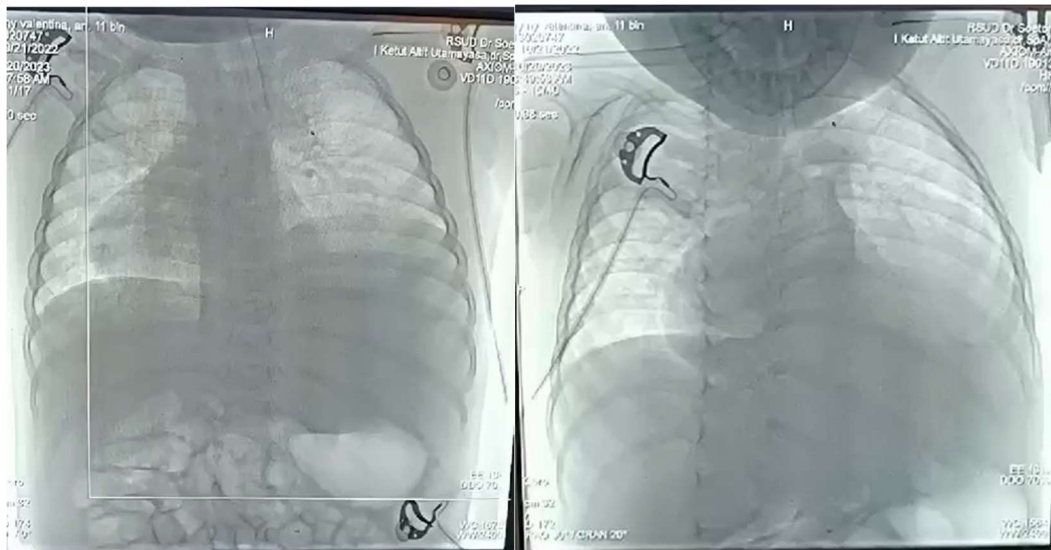


Figure 2: Fluoroscopy shows an eventration picture of the left diaphragm

The patient's lung condition improved gradually although oxygen saturation still fluctuated between 79-91%. An increase in FiO₂ was made to maintain oxygen saturation, but the patient still required periodic mechanical ventilation support. In addition, observation continued on hemodynamics, urine production, and fluid balance which showed fluctuations in positive and negative balances, indicating the need for strict fluid management. A decrease in leukocytosis from very high values without fever indicated an immune response to chronic infection or other causes such as oral candidiasis and diaper rash, which were treated with fluconazole and nystatin for candida infection and fusidate and hydrocortisone for rash.

On October 25, 2023, a decision was made to proceed with the surgery for plication of the sinistra diaphragm on October 26, 2023 because it was suspected that the eventration of the sinistra diaphragm affected the patient's respiratory condition, making it difficult to wean the patient off the ventilator. This was also confirmed by fluoroscopic examination where paradoxical movement of the left diaphragm was found (Figure 2).

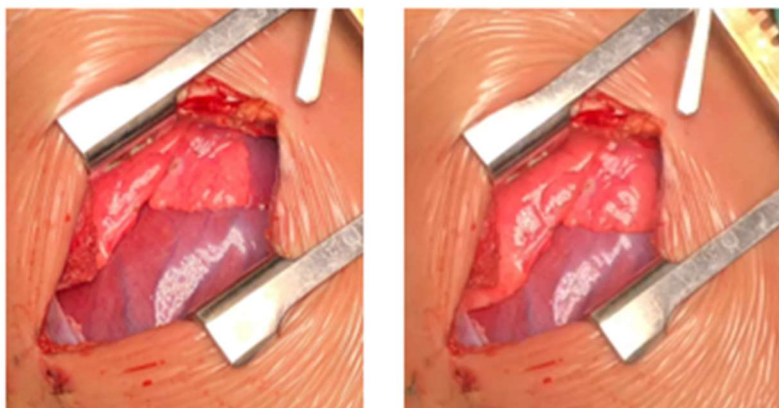


Figure 3: Left diaphragm dome as high as Intercostal space 4-5 (before plication)

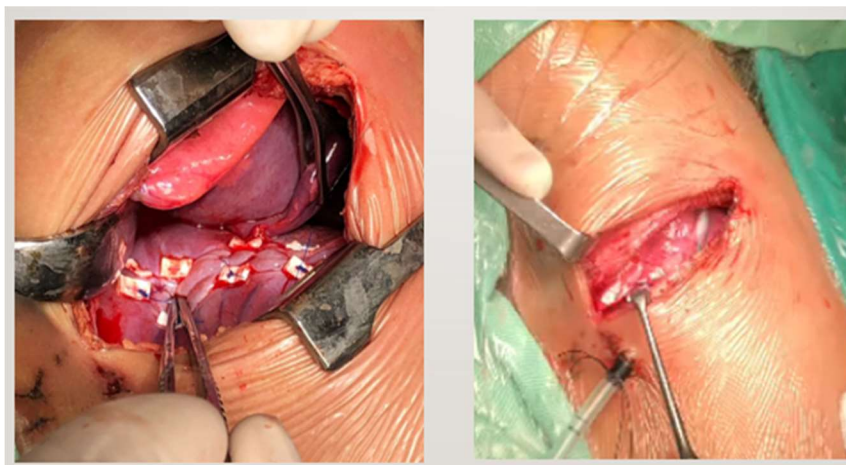
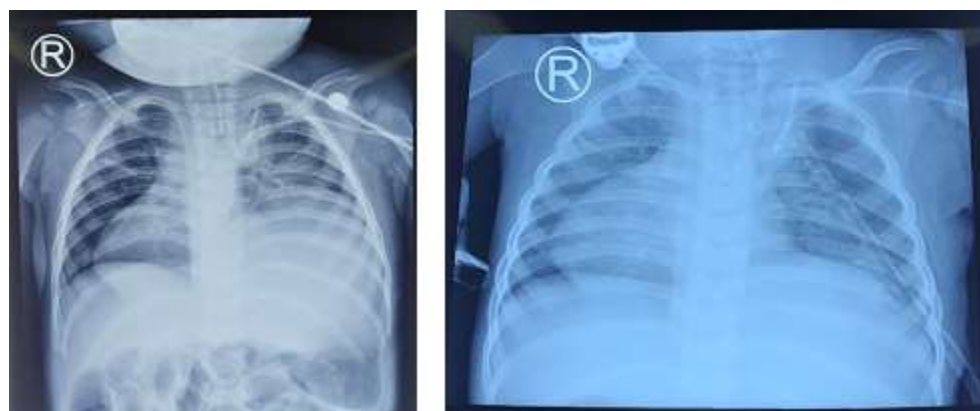


Figure 4: Post plication of left diaphragm and lung recruitment



24/10/2023

26/10/23

Figure 5: Chest x-ray before left diaphragm plication (Left), Chest x-ray after left diaphragm plication (Right)

Durante plication obtained left diaphragm dome as high as Intercostal space 4-5 and atelectasis in the inferior lobe of the left lung (Figure 3), then performed diaphragm plication with 4-0 polypropylene thread as many as 4 sutures with pledget, post-plication evaluation, diaphragm dome down, wash the thoracic cavum with warm NaCl 0.9%, and performed recruitment maneuver so that the inferior lobe of the left lung expands (Figure 4). Post operation left diaphragm plication, the patient showed significant improvement (Figure 5), although some

parameters still need to be closely monitored. In terms of ventilation, the patient experienced improvement in respiratory effort with the adjusted ventilation mode from PSIMV to SPONT although oxygen saturation was still fluctuating in the range of 80-90% and finally through tracheostomy because the patient had been tubed in on ventilator for almost 1 month. Chest movements were symmetrical but started with retractions when ventilator weaning was attempted and vesicular breath sounds were still heard despite rhonchi, indicating a large number of secretions and the patient fell into pneumonia and sepsis shock as she still needed adrenaline support and nor-adrenaline pump.

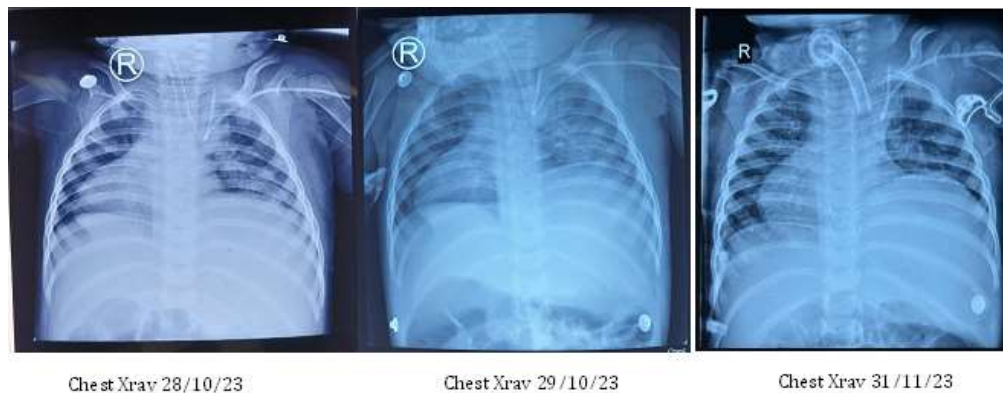


Figure 6: Chest x-ray evaluation showed left diaphragm plication failure

The patient also showed that 2-3 days post op plication, the left diaphragm had risen back to Intercostal space 4-5 (Figure 6). Many factors could have contributed to this, including suspicion of wound dehiscence at the plication suture due to sepsis. The next concern is also the delay in the decision to perform diaphragm plication early on so that the patient had already fallen into sepsis shock, making it more difficult to wean the ventilator.

Clinically, this patient showed various health problems, including respiratory acidosis, purulent secretions, and decubitus ulcers, indicating the need for more attention in critical care. Supportive therapy was also required, including ventilator adjustment, chest physiotherapy, and wound care for decubitus. Periodic hemodynamic and urine production monitoring reflected the seriousness of the patient's condition, where vital values showed marked fluctuations that could lead to further deterioration in clinical status.

During the treatment period, the patient experienced a series of events that led to a more severe condition, including desaturation and eventually cardiac arrest. The resuscitation process performed on the patient, although successful, could not prevent the patient from dying of shock sepsis with Multiple Organ Dysfunction Syndrome (MODS). The final decision to stop treatment by providing education to the family demonstrates transparency and emotional support which is important in end-of-life situations.

CONCLUSION

This study identified that diaphragm dysfunction, specifically diaphragm paralysis, is a frequent complication in patients after open heart surgery. This condition is closely related to phrenic nerve injury which can occur due to various factors, including the use of cold cardioplegia techniques. This injury to the phrenic nerve results in respiratory distress as seen by decreased oxygen saturation and increased respiratory effort in patients. To address this, diaphragmatic plication procedures are often applied, especially in children under one year of age, to improve respiratory function and speed up ventilator removal. The results of the treatment showed improved hemodynamic stability and improved pulmonary conditions, although the patient still required close postoperative monitoring.

To reduce the risk of phrenic nerve injury during open-heart surgery, the implementation of safer and minimally invasive surgical techniques is recommended. In addition, management of patients with this complication requires multidisciplinary collaboration between cardiac surgeons, critical care and

physiotherapists to ensure comprehensive care and optimal clinical outcomes. Regular assessment of the patient's respiratory function and hemodynamic status is crucial, which may include fluoroscopic evaluation or phrenic nerve stimulation, to facilitate the early identification of diaphragm dysfunction. Due to the effectiveness of diaphragm plication in improving postoperative respiratory dysfunction, this procedure can be considered as a standard therapeutic option in cases of diaphragm paralysis. With these measures, it is hoped that postoperative complications such as diaphragmatic dysfunction can be treated more effectively and earlier, thereby increasing the success rate of patient recovery.

REFERENCES

- [1] L. K. Nason, C. M. Walker, M. F. McNeeley, W. Burivong, C. L. Fligner, and J. D. Godwin, "Imaging of the diaphragm: anatomy and function," *Radiographics*, vol. 32, no. 2, pp. E51–E70, 2012, doi: 10.1148/rg.322115127.
- [2] T. H. Akay *et al.*, "Diaphragmatic paralysis after cardiac surgery in children: incidence, prognosis and surgical management," *Pediatr. Surg. Int.*, vol. 22, pp. 341–346, 2006, doi: 10.1007/s00383-006-1663-2.
- [3] Y.-B. Zhang, W. Xu, S.-J. Li, K. Yang, X.-D. Sheng, and Y. A. N. Jun, "Postoperative diaphragmatic paralysis after cardiac surgery in children: incidence, diagnosis and surgical management," *Chin. Med. J. (Engl.)*, vol. 126, no. 21, pp. 4083–4087, 2013, doi: 10.3760/cma.j.issn.0366-6999.20131173.
- [4] Z. Zachariou, *Pediatric Surgery Digest: Second Edition*. Springer Nature, 2022. doi: 10.1007/978-3-030-80411-4.
- [5] M. E. Höllwarth and P. Puri, *Pediatric Surgery: Diagnosis and Management*. Berlin, Heidelberg: Springer, 2006. doi: 10.1007/978-3-662-43567-0.
- [6] N. Najikhah and P. Maulidiana, "Factors Associated With Incidence Of Hypertension In The Patek Health Center Area Aceh Jaya District," *Transpublika Int. Res. Exact Sci.*, vol. 2, no. 1, pp. 8–14, Apr. 2024, doi: 10.55047/tires.v2i1.1135.
- [7] V. J. Aguirre, P. Sinha, A. Zimmet, G. A. Lee, L. Kwa, and F. Rosenfeldt, "Phrenic nerve injury during cardiac surgery: mechanisms, management and prevention," *Hear. Lung Circ.*, vol. 22, no. 11, pp. 895–902, 2013, doi: 10.1016/j.hlc.2013.06.010.
- [8] D. H. Parikh, A. W. Auldist, D. C. G. Crabbe, and S. S. Rothenberg, *Pediatric thoracic surgery*. London: Springer, 2009. doi: 10.1007/b136543.
- [9] S. Talwar, S. Agarwala, C. M. Mittal, S. K. Choudhary, and B. Airan, "Diaphragmatic palsy after cardiac surgical procedures in patients with congenital heart," *Ann. Pediatr. Cardiol.*, vol. 3, no. 1, pp. 50–57, 2010, doi: 10.4103/0974-2069.64370.
- [10] J. Lemmer *et al.*, "Postoperative phrenic nerve palsy: Early clinical implications and management," *Intensive Care Med.*, vol. 32, no. 8, pp. 1227–1233, 2006, doi: 10.1007/s00134-006-0208-4.