ISSN: 1607-8322, e-ISSN: 2220-5799 **Anaesthesia, Pain & Intensive Care**
Vol 27(2); April 2023 DOI: 10.35975/apic.v27i2.2177

CASE REPORT **PEDIATRIC ANESTHESIA**

Anesthesia and pain management in a pediatric patient with patent ductus arteriosus undergoing repair of recurrent diaphragmatic hernia

Reza Widianto Sudjud[1], Doddy Tavianto[2], Budiana Rismawan[3], Hana Nur Ramila[4], Muhammad Budi Kurniawan[5], Aria Annasya[6]

Author affiliations:

1. Reza Widianto Sudjud, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital, Bandung, West Java, Indonesia; E-mail: hreza.widianto.sudjud@unpad.ac.id ORCID: {0000-0001-5966-4846}
2. Doddy Tavianto, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital, Bandung, West Java, Indonesia; E-mail: dytavianto@gmail.com ORCID: {0000-0002-5724-9312}
3. Budiana Rismawan, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital, Bandung, West Java, Indonesia; E-mail: budianarismawan@gmail.com ORCID: {0000-0002-7323-5631}
4. Hana Nur Ramila, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital (, Bandung, West Java, Indonesia; E-mail: hananurramila@gmail.com ORCID: {0000-0003-3183-0210}
5. Muhammad Budi Kurniawan, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital, West Java, Indonesia; E-mail: meet_dbudi@yahoo.com ORCID: {0000-0001-5115-557X}
6. Aria Annasya, Faculty of Medicine Universitas Padjadjaran, Dr. Hasan Sadikin General Hospital, Bandung, West Java, Indonesia; E-mail: ariaannasya@gmail.com ORCID: {0000-0003-4520-2063}

Correspondence: Dr. Djoni Kusumah Pohan; **E-mail:** kakv.rshs@gmail.com **Phone:** +62 85278466279

ABSTRACT

Congenital diaphragmatic hernia is caused by a defect in the closure of the pericardio-peritoneal canal. It is a real challenge for the anesthetists during the perioperative management as serious complications including hypoxia and hypercarbia may threaten the life of the child. Mortality remains high due to associated pulmonary hypoplasia and pulmonary hypertension.

We present a case of a 15-month-old boy, known patient of recurrent congenital diaphragmatic hernia, scheduled to undergo laparotomy to repair his diaphragmatic defect. The patient presented with shortness of breath, and his chest X-ray revealed a diaphragmatic hernia. He was in optimal condition. General anesthesia was chosen with intubation. The patient remained stable and his laboratory tests were within normal limits. We discuss the associated problems during anesthesia and the optimum perioperative management.

Abbreviations: CDH - congenital diaphragmatic hernia; ECMO - Extracorporeal membrane oxygenation; ETT - Endotracheal tube

Key words: congenital diaphragmatic hernia, hypoxia, pulmonary hypoplasia

Citation: Sudjud RW, Tavianto D, Rismawan B, Ramila HN, Kurniawan MB, Annasya A. Anesthesia and pain management in a pediatric patient with patent ductus arteriosus undergoing repair of recurrent diaphragmatic hernia. Anaesth. pain intensive care 2023;27(2):280−284; **DOI:** 10.35975/apic.v27i2.2177

Received: August 28, 2022; **Reviewed:** November 05, 2022; **Accepted:** December 10, 2022

1. INTRODUCTION

Congenital diaphragmatic hernia (CDH) is a malformation characterized by a defect in the foramen of Bochdalek in the posterolateral part of the diaphragm where the abdominal viscera enter the thorax through the foramen during the fetal life. According to reports, CDH occurs in 1 in 4000 live births. The exact incidence of CDH is unknown because death often occurs in the early life and it is also known as a cause of mortality.

Approximately 50% of CDH is associated with malformations, especially of the heart, intestines, bones, and defects of kidneys. Diaphragmatic hernias are also associated with trisomy 13, 18 and 45 XO and have been reported as an abnormality in Goldenhar, Beckwith-Wiedemann, Pierre Robin, Goltz-Gorlin and Rubella syndromes.[1]

The location of CDH is most often found in the left hemithorax with a defect in the posterior diaphragm (Bochdalek foramen). In approximately 70% of infants, CDH can occur in the right hemithorax with defects in the anterior or posterior diaphragm. Some cases of CDH are asymptomatic which is common in Morgagni type.[1]

Congenital diaphragmatic hernia (CDH) that persists after surgical correction usually left breathing problems caused by pulmonary hypoplasia due to prior visceral organ compression. Patients with CDH after surgical correction may experience some disturbances such as growth disorders, eating disorders, failure to thrive, neurocognitive delay, hearing loss, behavior disorders in childhood and adolescence due to extracorporeal membrane oxygenation (ECMO) therapy.[2]

Congenital diaphragmatic hernia (CDH) has been known for years, but the survival after surgery remained poor until the 20th century. Currently the researchers of pediatric surgery report a very low mortality rate in CDH cases.[3]

2. CASE REPORT

We report a case of a one-year old boy who suffered from shortness of breath since 2 weeks of hospital admission. Written informed consent was obtained from his parents before we submitted this case report. There was no history of fever or having bluish lips. There were no complaints of additional breath sounds. The functions of defecation and urination were normal. Complaint of shortness of breath was first noticed when the patient was 10 months old. Because of his complaints, the patient was taken to Syamsudin Sukabumi Hospital and a chest X-ray was taken. It was found that the intestine had entered the chest cavity and a left CDH was diagnosed.

The patient underwent a diaphragmatic hernia repair operation in June 2021 at Syamsudin Hospital with intraoperative findings Morgagni type diaphragmatic hernia with ± 2 cm diameter, partial herniation of the ileum into the thoracic cavity, and closure of the defect without mesh. One month after surgery, the patient complained of recurrent shortness of breath. The patient then was brought to Assyifa Sukabumi Hospital and was hospitalized for 3 days. During the treatment, a chest X-ray and echocardiography were done. It was found that there was a patent ductus arteriosus and the intestine was again found in the chest cavity. The patient was given oxygen therapy 2 L/min via nasal cannula, nebulized with salbutamol and budesonide inhalation every 4 h, salbutamol powder 3 x 0.8 mg PO, inj. ceftriaxone 2 x 500 mg IV, captopril 3 x 3 mg IV, furosemide 2 x 10 mg IV, spironolactone 1 x 12.5 mg PO. He was referred to the Hasan Sadikin General Hospital for further treatment with a suspected Recurrent CDH and a hernia repair operation was planned.

On physical examination, his pulse rate was 100/min, respiration rate 26/min, SpO_2 98% pre ductal and 88% post ductal on room air. On thoracic examination, the shape and movements of thorax were asymmetrical and there was epigastric retraction. On lung examination, the breath sounds were more on right side than the left side, with no slime, bowel sounds were heard in the left hemithorax. Heart sounds were regular, with no murmur. On examination, the abdomen was found scaphoid, soft, not hyperemic, no gut contour, no gut twisted, and weak bowel sounds.

Laboratory examination on November 24, 2021 was within normal limits. On thoracic AP X-ray, it was found that the trachea was in the middle, heart was difficult to assess. The hilum was superimposed by the heart shadow, and the outline was difficult to assess. On the lateral chest X-ray, the anterior sinus was covered by an opaque shadow with sharp posterior sinus (Figure 1). The retrosternal space was bright with multiple lucency in the anterior hemithorax which appeared to be associated with the cavity. The impression was labeled as left diaphragmatic hernia. On abdominal radiology, it was found that the preperitoneal fat was clear, the psoas line was not clear, the contours of the two kidneys were not clear. The orogastric tube was seen attached with the distal end projecting at the level of the left L2−3 vertebra.

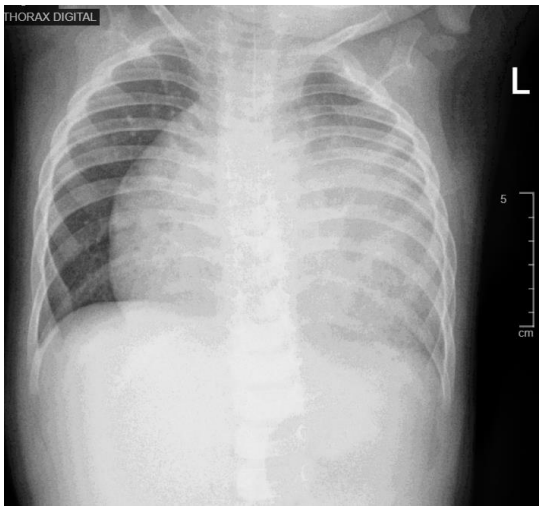

Figure 1: X-ray thorax; AP view

Patient was brought to the operating room and an IV line attached. A warming blanket was used on the operating table, anesthesia machine with pediatric Jackson–Rees circuit, anesthesia equipment, anesthetic drugs, and equipment for patient monitoring were prepared. An orogastric tube was passed to empty the stomach.

A precordial stethoscope was attached on the left chest. Induction of anesthesia was done with 100% O_2, and sevoflurane. Fentanyl 20 g (2 g/kg) was given for analgesia and the muscle relaxant atracurium 1 mg IV was given. Maintenance of anesthesia was done using oxygen, sevoflurane, fentanyl and intermittent atracurium muscle relaxant.

The surgery took about 2 h. During surgery, the heart rate (HR) remained stable between 110–130 beats/min and SpO_2 between 97–99%, with normal sinus rhythm. Dextrose + sodium chloride solution was infused @ 44 mL/h with a microdrip burette. Blood loss was estimated to be ± 20 ml, with urine output approximately 40 ml.

An anteromedial left diaphragm defect, measuring 2 x 2.5 cm was found. Intestine was anastomosed to transverse colon. Valsalva maneuver was performed when the left diaphragmatic defect was sewn. The right and left lungs expanded after closing the defect.

After the operation was done, erector spinae block (ESP) was performed using 1 mL of 0.5% bupivacaine (Figure 2).

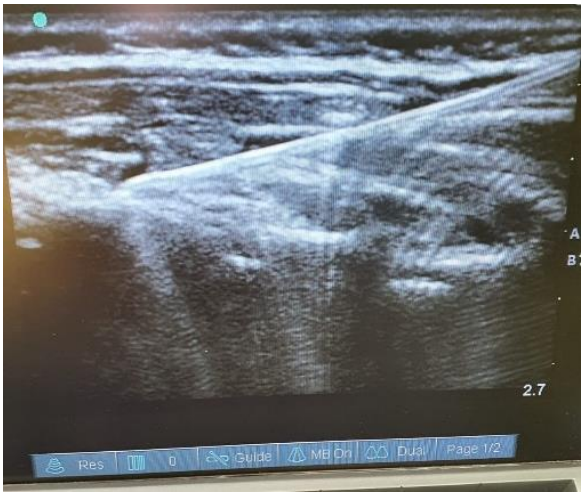

Figure 2: ESP block

Extubation was done when fully conscious and he was transferred to the PICU for postoperative care. Postoperative chest X-ray showed both lungs were inflated (Figure 3). On the third postoperative day, the patient was transferred to the ward and was discharged on 5th postoperative day.

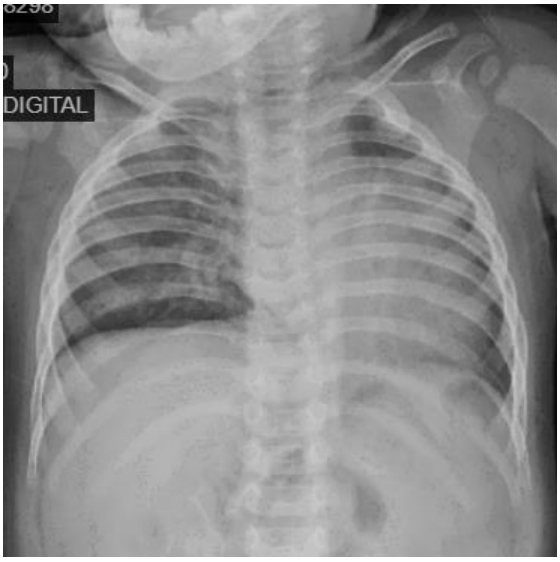

Figure 3: Anteroposterior X-ray thorax

3. DISCUSSION

Diaphragmatic hernia generally occurs together with other abnormalities in 20−50% of cases. It can be seen as a non-chromosomal syndrome (mental retardation, cleft lip and heart defects) or as a chromosomal defect (trisomy or non-trisomy). Congenital anomalies were the most common cause of neonatal death (1.7 per 1000 births) and diaphragmatic hernia was accountable for approximately 4−10% of deaths. Gestational age at the time of herniation could determine the degree of pulmonary hypoplasia, reflecting the severity of lung pathology and life expectancy, which usually varies widely from 25% to 83%.[4]

The ipsilateral lung is generally compromised and herniation of the bowel can compress and delay the maturation of both lungs. Diaphragmatic hernias are usually exacerbated by pulmonary hypertension and are associated with 40-50% mortality. Cardiopulmonary compromise generally occurs primarily due to pulmonary hypoplasia and hypertension, rather than the mass effect of herniated viscera. Pulmonary hypoplasia and persistent pulmonary hypertension are two important factors determining the final outcome. Other factors are associated congenital and chromosomal abnormalities.[4]

Early antenatal diagnosis with appropriate perioperative management is the main thing determining a good prognosis. Nitrous oxide (N_2O) should be avoided for it would be diffusing into the viscera and increase pulmonary compression. Problems that might arise in patients with diaphragmatic hernia are respiratory distress related to pulmonary parenchymal hypoplasia,

hypoxemia, hypercarbia, acidemia, metabolic acidosis, pulmonary hypertension, right-to-left shunting, malrotation (may occur in > 50−100% of patients), and the occurrence of hypoglycemia and hypothermia.

Based on physical examination, our patient showed a cardiac abnormality with the impression of a PDA L−R Shunt, although he was stable without inotropic drugs.

Emergency surgery was the standard approach to diaphragmatic hernias during the 1980s for it was believed that hernia reduction would improve respiratory status, allowing the lungs to re-expand. When it was found that the lungs were hypoplastic, not atelectasis and the arteriolar muscles were impaired which caused pulmonary hypertension, the delay of initiation of surgery had been introduced. It was reported that delayed repair of a diaphragmatic hernia for 24 h had the rationale that the risk of pulmonary hypertension was reduced. In 1986, Cartlidge et al. stressed upon preoperative stabilization. Sakai et al. stated that lung compliance was still impaired after repairment.[2,5,6,7]

Shonbogue et al. reported that the life expectancy of critically ill neonates reduced to 58% when ECMO was used, by resting the lungs and possibly reduced the risk of pulmonary hypertension. The relatively short stabilization period might be inadequate in hemodynamically labile patients. Langer et al. also concluded that diaphragmatic hernia repair should be performed only when the patient was sufficiently stable.

Ideally, invasive circulation monitoring is desirable. However, it was not performed in this patient due to patient's stable condition without inotropic support and no signs of pulmonary hypertension were found with minimal bleeding probability. Intraoperative management should ensure adequate gas exchange, proper monitoring and electrolyte replacement, aspiration prevention, and control of perioperative pain.

At the time of induction, the main concern was the possibility of a full stomach, so it was necessary to insert an orogastric tube (OGT) before induction and aspirate. If the infant has not been intubated, rapid sequence induction (RSI) or awake intubation (in predicted difficult intubation) without bag and mask ventilation is desirable to prevent abdominal overdistention and herniation across the midline. Our patient had ETT and OGT inserted. Anesthesia in newborns with diaphragmatic hernias is recommended using general inhalation anesthesia with ventilation control. Nitrous oxide should be avoided to prevent diffusion into the intestinal lumen so that intra-thoracic and intra-abdominal bowel expansion can be prevented. In most cases, a combination of muscle relaxants and oxygen-opioids are needed. Anesthetics that depress the myocardium should be avoided until the chest is decompressed.[10,12]

Controlled ventilation is guided by arterial carbon dioxide tension ($PaCO_2$) and use of muscle relaxants. Atracurium 0.5 mg/kg was used. Other drugs such as rocuronium, vecuronium, and mivacurium have not been reported in neonates. [10,12]

In infants, heat loss in a cold environment could easily occur because of the relatively larger body surface area and the lack of subcutaneous fat that functions as a heat barrier. Heat loss from the body surface can be minimized by increasing the operating room temperature, using a heating blanket, and warming and humidifying anesthetic gases.[12]

After the surgery was done, the mucus in the nasal and oral cavities were cleaned. The anesthetic was stopped and 100% oxygen administered for 5−15 min. Postoperatively, the patient was transported to the PICU.

4. CONCLUSION

Early antenatal diagnosis, avoidance of high airway pressures during ventilation and hemodynamic stability can lead to good outcomes in patients with congenital diaphragmatic hernia. Patients with diaphragmatic hernias experience respiratory problems with non-expansive lungs, the possibility of pneumothorax, and the possibility of pulmonary hypoplasia. Anesthesia in newborns requires special equipment and close monitoring, as well as more careful preparation. Proper airway management with controlled ventilation during surgery, strict fluid balance assessment and prevention of hypothermia are important to be closely monitored.

Postoperatively, close monitoring of respiratory function and provision of mechanical ventilation is required. Clinical monitoring with serial blood gas and radiological analysis are carried out to assess the development of postoperative respiratory function.

5. Conflict of interest

None declared by the authors.

6. Authors' contribution

All authors took part as a team in concept, organizing the study, re-evaluation of the study, language and literary corrections, literature review, evaluation and conclusion of findings, drafting the manuscript

All authors have read the manuscript and endorse it.

7. Patient's consent

Written permission was obtained from the father of the child to publish this report.

8. REFERENCES

1. Merin RG. Congenital diaphragmatic hernia: from the anesthesiologist's viewpoint. Anesth Analg. 1966 Jan-Feb;45(1):44-52. [PubMed]
2. Van Meurs K, Lou Short B. Congenital diaphragmatic hernia: the neonatologist's perspective. Pediatr Rev. 1999 Oct;20(10):e79-87. [PubMed] DOI: 10.1542/pir.20-10-e79
3. Keijzer R, Puri P. Congenital diaphragmatic hernia. Semin Pediatr Surg. 2010 Aug;19(3):180-5. [PubMed] DOI: 10.1053/j.sempedsurg.2010.03.001
4. Shanbhogue LK, Tam PK, Ninan G, Lloyd DA. Preoperative stabilisation in congenital diaphragmatic hernia. Arch Dis Child. 1990 Oct;65(10 Spec No):1043-4. [PubMed] PMCID: PMC1590261 DOI: 10.1136/adc.65.10_spec_no.1043
5. Nagata K, Usui N, Kanamori Y, Takahashi S, Hayakawa M, Okuyama H, Inamura N, Fujino Y, Taguchi T. The current profile and outcome of congenital diaphragmatic hernia: a nationwide survey in Japan. J Pediatr Surg. 2013 Apr;48(4):738-44. [PubMed] DOI: 10.1016/j.jpedsurg.2012.12.017
6. Cartlidge PH, Mann NP, Kapila L. Preoperative stabilization in congenital diaphragmatic hernia, Arch Dis Child. 1986 Dec;61(12):1226-8. [PubMed] PMCID: PMC1778183 DOI: 10.1136/adc.61.12.1226
7. Sakai H, Tamura M, Hosokawa Y, Bryan AC, Barker GA, Bohn DJ. Effect of surgical repair on respiratory mechanics in congenital diaphragmatic hernia. J Pediatr. 1987 Sep;111(3):432-8. [PubMed] DOI: 10.1016/s0022-3476(87)80475-4
8. Langer JC, Filler RM, Bohn DJ, Shandling B, Ein SH, Wesson DE, Superina RA. Timing of surgery for congenital diaphragmatic hernia: is emergency operation necessary? J Pediatr Surg. 1988 Aug;23(8):731-4. [PubMed] DOI: 10.1016/s0022-3468(88)80413-5
9. Chatterjee D, Ing RJ, Gien J. Update on Congenital Diaphragmatic Hernia. Anesth Analg. 2020 Sep;131(3):808-821. [PubMed] DOI: 10.1213/ANE.0000000000004324
10. Badillo A, Gingalewski C. Congenital diaphragmatic hernia: treatment and outcomes. Semin Perinatol. 2014 Mar;38(2):92-6. [PubMed] DOI: 10.1053/j.semperi.2013.11.005
11. Haroon J, Chamberlain RS. An evidence-based review of the current treatment of congenital diaphragmatic hernia. Clin Pediatr (Phila). 2013 Feb;52(2):115-24. [PubMed] DOI: 10.1177/0009922812472249

