

## Anesthetic Management of Conjoined Twins Separation in A Remote Area of Indonesia: a case report

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### Abstract

**Introduction:** Conjoined twins is a rare condition which brings various complications to the patients since the early life. Careful management needs to be undertaken to ensure patients' safety. Multidisciplinary medical team and advanced medical instruments must be prepared for the pre-surgery, separation surgery, and post-surgery management.

**Objectives:** This case report aims to provide valuable insights into the complexities of managing conjoined twins in challenging circumstances, emphasizing the need for careful planning, teamwork, and specialized anesthesia care to achieve positive outcomes.

**Case:** Here, we present a case of conjoined twins in a remote area of Indonesia. Both patients were both in a critical condition while patients transport were not possible, thus emergency separation was obliged in the remote hospital.

**Results:** Multidisciplinary medical team and medical instruments were dispatched from larger cities. Several modifications were required to adapt to the lack of infrastructure in the remote area. Conjoined twins were successfully separated.

**Conclusions:** This proves that careful anesthetic management and teamwork of multidisciplinary team could contribute to a successful separation surgery of conjoined twins.

**Keywords:** anesthetic management, conjoined twins, separation surgery.

### 1. Introduction

Conjoined twins are extremely rare, with estimated prevalence ranging once in 50,000 to 100,000 births (Mutchinick et al., 2011). Higher incidence has been observed in Southeast Asia and India with occurrence ranging once in 14,000 to 25,000 births (Hanson, 1975; Spitz, 1996). Conjoined twins mostly are female (Mian et al., 2017). In United States, conjoined twins have poor prognosis, with overall morbidity around 62% and 70% during the first 48 hours (Willobee et al., 2019). Pre-surgery intensive care and collaborative team is fundamental so that conjoined twins could survive post separation.

Conjoined twins are classified into several classification based on the involved body region, i.e., thoracopagus, omphalopagus, pygopagus, and ischiopagus (Brock & Johnson, 2021). In the omphalopagus conjoined twins, fused liver is common. Separation surgery of the liver is very challenging; thus, it is classified as a high-risk procedure because liver which is known as a highly perfused organ and intra-operative blood loss that would likely occur is a critical determinant of the infants' separation surgery.

Here, we present a case of an omphalopagus twins (conjoined twins sharing part of gastrointestinal system and abdominal wall), with sepsis, diaphragmatic hernia, and neonatal icterus. This case itself was difficult because it was found in a remote area, Larantuka, where there were very limited medical facility and resources. Larantuka itself is a sub-district in East Flores, East Nusa Tenggara Province, Indonesia. This small town area is 75.91 km<sup>2</sup>, with a population in 2020 of around 45.515 people. It takes 4 hours driving to the nearest bigger city Maumere, and 1 hours flight to Kupang, the capital city of East Nusa Tenggara which located in a different island.



## 2. Objectives

The objectives of this case report on the "Anesthetic Management of Conjoined Twins Separation in a Remote Area of Indonesia" are to underscore the intricate challenges faced in handling conjoined twins, especially in resource-limited and remote settings. It aims to highlight the pivotal role of a multidisciplinary medical team and meticulous planning in navigating the complexities of pre-surgery, separation surgery, and post-surgery care. The report emphasizes the critical contribution of anesthesiologists and anesthesia management in achieving a successful separation, particularly in high-risk procedures like liver separation. It underscores the necessity of effective communication and collaboration among medical professionals, local authorities, and stakeholders in remote regions with limited medical infrastructure. Additionally, the report accentuates the need for tailored surgical techniques and meticulous preparation for each twin, while also stressing the significance of ongoing post-surgery monitoring and intensive care.

## 3. Case

A pair of female conjoined twins were born on 12.15 A.M local time (GMT +8) on April 28<sup>th</sup> 2023. This case was found in a hospital in East Flores, East Nusa Tenggara, Indonesia, which is a small remote island, also located very far from any big city. They were born by caesarean section with general anesthesia at 39 weeks gestation, this is a second pregnancy of the mother, the first born is three years old now and was spontaneously delivered. Total birth weight was 4,300 grams, with the first baby being smaller than the second one. Their mother has no chronic medical condition, does not use tobacco or alcohol, got routine antenatal care and had twice obstetric ultrasound examinations but never diagnosed with conjoined twins through the entire pregnancy. The first baby (next will be labeled as the red baby) had APGAR score of 3 at the first minute and 7 at the fifth minute. She had a respiratory distress with Down Score of 5, then moved to Neonatal Intensive Care Unit (NICU) using infant T-piece resuscitator Neopuff™ (Figure 1). In the NICU, the patient was intubated using no.3 non-cuffed endotracheal tube and Pressure Control-Continuous Mandatory Ventilation (PC-CMV) mode on ventilator. The second baby (next will be labeled as the green baby) was born with APGAR score of 4 at the first minute and 8 at the fifth minute and also experienced respiratory distress, with Down Score of 1 – 2. She was then transferred to NICU with 0.5 litre per minute of oxygen using nasal cannula. Afterwards, the green baby condition improved and oxygen supplementation was taken off after several hours.

CT scan examination (Figure 2) demonstrated a diaphragmatic hernia with intestinal organ protruding to the thoracic cavity and pushed mediastinum to the right side and caused left lung collapse on the red baby. There was an abdominal wall defect with partial liver fusion between the twins. Liver fusion was also confirmed by ultrasound examination. Both patients were diagnosed with early onset neonatal sepsis. Intravenous ampicillin 108 mg b.i.d and intravenous amikacin 16 mg b.i.d were given.



**Figure 1. The twins during transport to NICU. The red baby (left) was on T-piece resuscitator, while the green baby (right) was on nasal cannula**

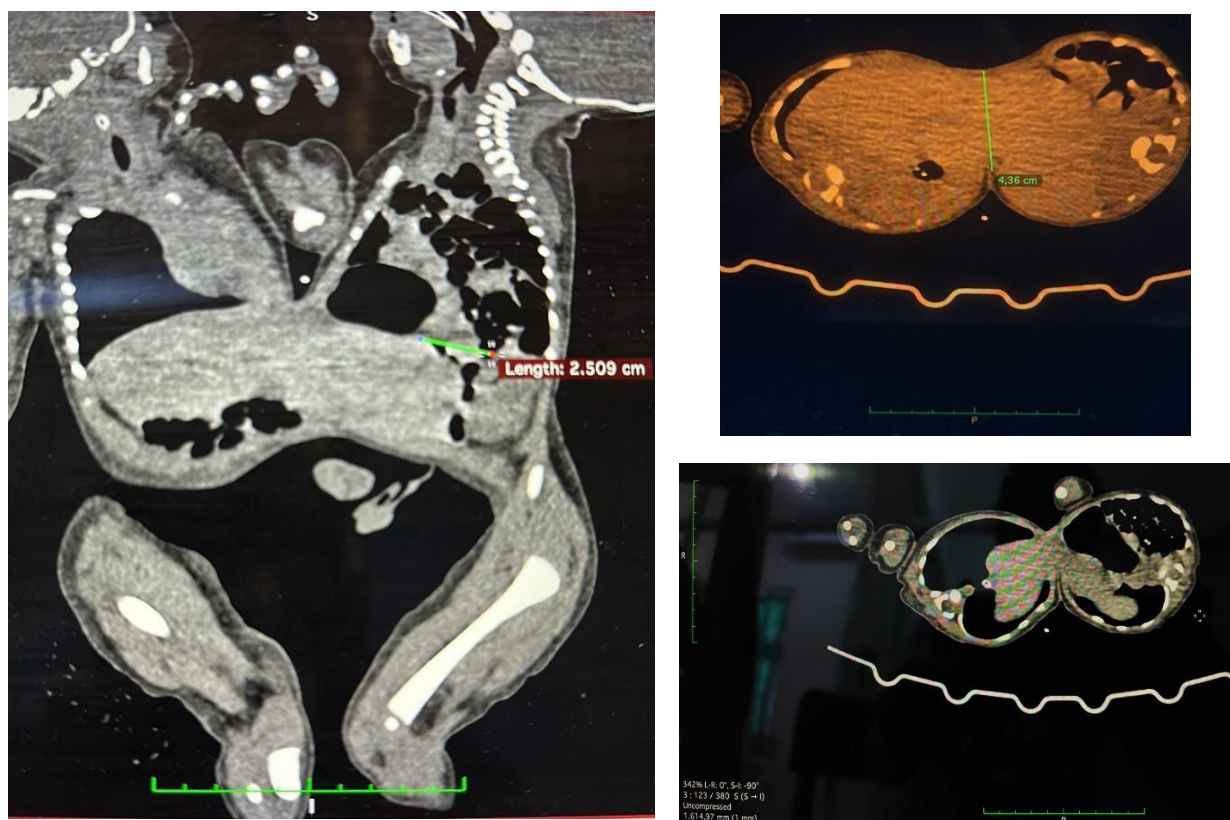


#### 4. Results

##### a. Pre-surgery care and preparation

We held two online conferences, there were attended by all of the medical staffs and pharmacist from surrounding medical centers; i.e Larantuka, Maumere, Kupang, and Surabaya. Larantuka local governors and stakeholders also joined the online meeting. Through the meeting, we communicated and listed all the instruments available at the center; and those unavailable ones that need to be provided from outside the center. Other than medical instruments, general planning was also discussed during the online meeting. There were two teams for the surgery, the initial team who came first (two days before the surgery) and the main team who came later. The initial team evaluated the general condition of the patients, they also decided the status of the surgery if it was going to be elective, urgent, or emergency surgery, and whether the patients were transportable to any of the bigger centers; i.e Kupang, Maumere, or Surabaya. The main team would arrive later to perform the surgery together with the initial team.

On their 5<sup>th</sup> day, the red baby had a severe desaturation with oxygen saturation as low as 68%, severe chest retraction, respiratory rate was 59 per minute, capillary refill time (CRT) <3 seconds, and heart rate was 150 bpm. Endotracheal tube (ETT) was found to be pulled off and Down score was 2. ETT was put back with non-cuff ETT number 3.0, 8 cm depth, connected to ventilator. The setting of ventilator was Pressure Control – Assist Control (PC-AC) mode, Fraction of Inspired Oxygen (FiO<sub>2</sub>) was 50%, respiratory rate was 60/min, Pressure of Inspired Oxygen (PIP) was 25 cmH<sub>2</sub>O, and Positive End-Expiratory Pressure (PEEP) was 5 cmH<sub>2</sub>O. She showed signs of jaundice, with Kramer score of 3 – 4. Direct bilirubin was 0.36 mg/dL and total bilirubin was 17.7 mg/dL. The patients were given phototherapy for a total of 24 hours in 2 separate days to treat the hyperbilirubinemia for 24 hours in 2 days.



**Figure 2.** CT scan examinations showing liver fusion between the twins

On 7<sup>th</sup> day, respiratory distress improved in the red baby (age 7 days old). Down score was 0 – 1, respiratory rate was 68/min, improved chest retraction, heart rate was 130 – 140 bpm, oxygen saturation was between 88 – 96%.



ETT number 3.0 was administrated with ventilator settings of PC-AC mode,  $\text{FiO}_2$  was 25%, respiratory rate was 60/min, PIP was 25  $\text{cmH}_2\text{O}$ , PEEP was 5  $\text{cmH}_2\text{O}$ , inspiratory time ( $T_i$ ) was 0.4, inspiratory to expiratory ratio (I:E) was 1:1.5, tidal volume was 6.4 ml. Patients experienced fever with temperature of  $37.9^\circ\text{C}$  –  $38.2^\circ\text{C}$  and  $38.20^\circ\text{C}$  –  $39.2^\circ\text{C}$  at night, respectively in the first and second baby. Each patient was given intravenous paracetamol 25 mg every 6 hours. Phototherapy was not effective due to the fever. By 6<sup>th</sup> July 2023 the initial team decided that the separation surgery was going to be an urgent one. They realized the risk of the green baby condition would deteriorate if the surgery got postponed even longer.

Prior to separation surgery, multidisciplinary medical team gathered and did a simulation of the separation surgery sequences. Then, the operating rooms (OR) were prepared according to the planned layout (Figure 3). Instruments and monitor were set based on the surgery plan. Operating table was substituted with infant warmer (Figure 3). Operating room 1 and 2 face each other.



**Figure 3. Operating room for conjoined twins separation. (A) OR #1 for the red baby. (B) OR #2 for the green baby. (C) OR #1 after setting. (D) OR #2 during preparation. (E) Operating table was substituted with infant warmer**



The medical team comprised of 2 anesthesiologists, 3 pediatric surgeons, 1 neonatologist, 2 pediatricians, 2 plastic surgeon, 2 anesthesiology residents, and a few nurses of whom were all dispatched from referral hospitals, except the anesthetic nurse. The hospital provided operating rooms, large oxygen cylinders, an observational monitor, 2 anesthetic machines, ultrasonography device, syringe pumps, anesthetic nurse and surgical instruments. Additional required instruments were hauled from hospitals in the capital city Surabaya, Kupang, and Maumere, such as an observational monitor, intra-arterial blood pressure catheter and device, central venous catheter (CVC) set, endotracheal tubes in 2.5 – 4 sizes, surgical instruments for conjoined twins neonates, harmonic surgical scalpel, Abbott© i-STAT 1 Portable Point of Care Analyzers to measure arterial blood gas analysis. Due to the lack of medical instruments, we used vaginal ultrasound probe to perform echocardiography and a linear ultrasound probe to insert the intra-arterial blood pressure catheter.



**Figure 4. Marking of liver fusion**

**b. Anesthesia induction and maintenance**

Separation surgery was performed on May 7th 2023 from 10.00 AM to 08:00 PM local time (GMT+8), the conjoined twins were 10 days old. The first and second baby was labeled as red and green baby, respectively. Incision mark was made to locate the position of the liver fusion (Figure 4). The patients were supposed to be ventilated using idle Bain circuit, however, because it was not available, we used modified type-F Mapleson with increased flow. No premedication was administered. First, we intubated the green baby, then the red baby. Green baby was in supine position, preoxygenated using 8 lpm flow, anesthesia induction using 0.3 mg of intravenous midazolam, 5 microgram of intravenous fentanyl, and inhaled sevoflurane. Two miligrams of rocuronium muscle relaxant was added. Anesthesia was maintained using inhaled Sevoflurane 2.2 minimum alveolar concentration (MAC). Patients were intubated using ETT number 4.0 non-cuff and they were well ventilated. Red baby was preoxygenated with 8 litres per minute (lpm) flow, anesthesia induction using Sevoflurane inhalation, two miligrams of rocuronium muscle relaxant was added. Central vein catheter (CVC) number 3.0 were secured in both patients. After several minutes of induction, the red baby oxygen saturation dropped from 96% to 30%, had bradycardia with the heart rate of 34 bpm, we checked the airway and breathing, the tubes were all in place, but deep chest retractions were seen, breathing looks heavier and breath sounds were decreased. We gave ventilation with PEEP, we administered 0,01 mg of intravenous epinephrine twice. Then the heart rate and saturation increased to normal.



c. Separation and reconstruction surgery

The twins were positioned in the operating table, heads and hands were covered with plastic, and surgery field was draped. Red baby on the left side and green baby on the right. The separation lasted from 10:00 AM to 1:00 PM local time (GMT+8). After separation, red baby stayed in the OR #1, while green baby was brought to the operating room #2 (Figure 5). After the separation, we found that the red baby got a left diaphragm defect at least 4 cm-wide, which caused the stomach, pancreas, and some parts of small intestines filled the left hemithorax, which also led to hypoplasia of the left lung. Both babies then had reconstructions of the abdominal wall by pediatric surgeons and the post-separation defect was closed by plastic surgeon. Final evaluation of the patients' post-surgery revealed free airway, controlled breathing with oxygen saturation of 97 – 98% with ventilator support. Then, patients were transferred to neonatal intensive care unit (NICU) and monitored. Alimentation started when patients were completely awake and not vomiting.



**Figure 5. Green baby was transferred from OR 1 to OR 2 after separation process was completed. (A) The process of green baby transfer. (B) The alley heading to the NICU from operating room. (C) NICU entrance gate**

d. Post-surgery management

One day post-surgery evaluation showed that the red baby was still in respiratory distress with occasional desaturation (oxygen saturation 81 – 100%). Temperature was 37.5° C. Jaundice was visible. Chest X-ray demonstrated lung infiltrates suggesting pneumonia. Post-surgery bloodwork revealed WBC of 15,200/ $\mu$ l. Ventilator support was given with settings Pressure Control-Bi-Level Positive Airway Pressure (PC-BIPAP), ETT number 3.0, FiO<sub>2</sub> was 90%, respiratory rate was 50/ minute, PIP was 20 cmH<sub>2</sub>O, PEEP was 5 cmH<sub>2</sub>O, Ti was 0.55, I:E ratio was 1:1.2. Intravenous Cefotaxime 112.15 mg b.i.d and intravenous Amikacin 16.8 mg b.i.d were administered. Intravenous Metamizole 50 mg t.i.d was given. The green baby showed no respiratory distress and on the first day of post operation was sedated and supported by ventilator. The next day green baby was extubated and direct breastfeeding could be initiated right afterwards.

## 5. Discussion

Based on the involved body regions, most common cases of conjoined twins are classified into 5 major classifications, (1) thoracopagus with the involvement of trunks, (2) omphalopagus, abdominal wall and possibly its organs are fused, (3) pyopagus where sacrum is involved, (4) ischiopagus is the fusion of hip, and (5) craniopagus is the fusion of the heads. Thoraco-omphalopagus is the most common conjoined twins with 28% prevalence, followed by thoracopagus as the second most common one with 18.5% prevalence (Kaufman, 2004). Less common conjoined twins are cephalopagus (fusion from head to umbilicus), and rachipagus (fusion from vertebral column) (Baken et al., 2013). Parapagus twins is defined as the fusion of lateral side of the body involving diaphragm to pelvis, with variable fusion of thorax and/or cranium. Parapagus is later categorized into parapagus diprosopus (extensive fusion of cranium and trunk, with two separate faces facing opposite direction);



and parapagus dicephalus (twins with the fusion starts from trunk and below, with separate heads and faces) (Mian et al., 2017).

Separation of conjoined twins is a very long procedure and have a high bleeding risk, thus, surgery during neonatal period is not recommended (Sato et al., 2021). Elective separation surgery of CT is recommended to take place between 2 – 4 months age (Spitz & Kiely, 2002). However, due to several life-threatening conditions such as stillborn, intestinal obstruction, omphalocele rupture, circulatory or respiratory failure, or urinary obstruction, separation surgery could be executed in an urgent or emergency settings (Shi et al., 2006). In this case, due to a diaphragmatic hernia in one of the babies, her left lungs were compressed causing atelectasis, which progress into respiratory distress, therefore emergency separation is demanded. The hospital where the babies were born was a small hospital in a remote area, where the facilities and medical staffs were limited. The only possible mode of transport to the nearest adjacent referral hospital was by aircraft and it was not feasible due to progressive deteriorating conditions of the conjoined twins.

A multidisciplinary team with good coordination and management was required for most of medical decisions taken to optimize pre-surgical conditions for babies. Before separation surgery took place, preparations were divided into resuscitation, stabilization, diagnosis, and definitive work-up (Utariani, 2020). In these patients, they both experienced respiratory distress. The red baby had more severe breathing problem, so she was intubated and put on the ventilator. The green baby was initially supported with low flow oxygen using nasal cannula then improved during the first three day, therefore oxygen supplementation was no longer needed. Both of them were monitored in the NICU. After being stabilized, patients underwent several imaging and laboratory examinations to support diagnosis. Abdominal CT scan revealed diaphragmatic hernia on the red baby and partial liver fusion between twins. Additionally, pneumonia (as a form of infection) and organ dysfunction (respiratory distress) was found, which support early onset neonatal sepsis diagnosis. Treatment was prescribed accordingly such as ventilation support and antibiotic.

Preparations and pre-surgery simulation are crucial in conjoined twins separation surgery. With only 60% of cases survive post-separation, therefore comprehensive management from multidisciplinary team is required in these cases (Brizot et al., 2011). Multidisciplinary team including pediatric surgeons, pediatric anesthesiologist, neonatologist, and nurses are required to perform this surgery (Ramlan et al., 2023). It requires 2 anesthetic teams, each team being assigned distinctive individual baby labeled with different colors (Utariani, 2020). The team was comprised of an anesthesiologist, anesthetic set, medications, and nurse anesthetists. However, it is rather difficult to compose this team in a remote area due to the lack of experienced specialist and licensed medical staffs in handling these cases. Unstable patients in critical conditions should not be transferred; therefore, emergency surgery was decided to be done in the same hospital where the patients are. In our case, additional team from bigger centers were dispatched to execute the surgery. Limitations in medical supplies and instruments were addressed by transporting them from adjacent bigger centers. After the team gathered, they planned and rehearsed the surgery in detail from receiving the patients in NICU until post-op recovery period. Intra-operative simulation was reviewed in details by all specialists involved in the surgery.

Before any anesthesia procedure started, patients were put on monitors. Each monitor was labeled using different color assigned for each baby to enable the distinction of patients and prevent any misrecognition; i.e. red label for first baby and green label for second baby. Performing intubation on conjoined twins with one baby on top of each other is not recommended nor ideal, as the baby on top could become pale and hypoxic, the baby below could suffer from excessive fluid buildup caused by gravity effect. Based on the experts' recommendation, intubation is preferred in a slightly tilted position, using cushion as a support to facilitate the ideal position (Utariani, 2020). It is suggested that the anesthesia used for intraoperative induction is inhaled Sevoflurane (Utariani, 2020). Anesthesia could be maintained using combinations of inhaled anesthesia and opioid given in intravenous anesthesia. Muscle relaxants commonly used are Rocuronium, Vecuronium, or Atracurium. Among other types of conjoined twins, omphalopagus is easier to separate (Aoun Tannuri et al., 2013). As long as the hearts are not fused, technically the separation could be done. Omphalopagus is the conjoined twins with the highest rate successful separation (Ramlan et al., 2023).



After separation surgery, patients are still intensively monitored in the NICU/ICU. Multidisciplinary medical teamwork is still required in this stage. Complications after separation surgery of CT is as complex as the separation procedure. Several potential problems that may be faced during post-surgery period are hypoventilation due to the remaining anesthetic agent, hypothermia due to long period of surgery and the cold temperature of operating room, laryngeal edema caused by long period of intubation, metabolism and circulation dysfunction, inflammation and infection response which may lead to sepsis, surgery wound that takes long time to heal, nutritional and psychic problem, and the effect of separation of the previously fused organ (Utariani, 2020).

## 6. Conclusion

Hereby, we presented a successful case of anesthesia management of conjoined twins separation in a remote area of Indonesia where the infrastructure and medical resources are limited. In conclusion, anesthesia management of similar cases, requires comprehensive management and detailed preparation to ensure better outcome.

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