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Case report

SUCCESSFUL ANESTHETIC MANAGEMENT FROM SEPARATION SURGERY OF PYGOPAGUS CONJOINED TWIN; LESSON-LEARNING WITH A TELEANESTHESIA

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ABSTRACT

Introduction: The management of conjoined twins requires multidisciplinary teamwork. The complex problems in conjoined twin separation surgery are challenging for anesthesiologists without experience in the management of conjoined twins. **Objective:** To describe anesthetic management and utilization of teleanesthesia in conjoined twin separation surgery. **Case Report**: Sixty days-old pygopagus type conjoined twins, with a total body weight of 7030 grams. Both babies looked healthy, moved actively, found no respiratory function disorders, were hemodynamically stable and had no congenital abnormalities. The sacral region's computerized tomography scan (CT-scan) reveals conjoined twins with skin unification and subcutaneous in the perianal region and no internal-vertebral-spinal fusion. Two anesthesia teams performed the management of anesthesia. After confirming there was no cross-circulation with the atropine test, we alternately induced anesthesia by inhalation technique while maintaining spontaneous breathing. Anesthesia was maintained with sevoflurane 2.0-3.0 vol%, in a mixture of oxygen and air with a flow of 4 L/min using Jackson Reese. Circulating volume, hemodynamic stability, and normothermia were maintained intraoperatively. The separation surgery lasted 20 minutes, and the total surgical time for each baby was two hours. Awake extubation was performed immediately after the surgery was complete. Both babies underwent postoperative care at the PICU and were discharged on day 11. During the pre-operative for surgery, the local team conducted telemedicine consultations with the pediatric anesthesia team at Dr. Soetomo hospital and performed intra-anesthesia telementoring. **Conclusion:** Careful preparation and pre-operative evaluation, proper intra-anesthesia maintenance and monitoring, as well as good communication and teamwork, are keys to successful anesthesia management in conjoined twin separation surgery. Consultation and assistance from an experienced team during surgery using teleanesthesia are significantly beneficial to the anesthesiologist without experience in conjoined twin separation surgery.

Keywords: Conjoined Twin; Childbirth Complications; Pygopagus; Telemedicine, Teleanesthesia

ABSTRAK

Pendahuluan: Penanganan bayi kembar siam memerlukan kerjasama tim dari berbagai multidisiplin. Kompleksitas permasalahan yang dapat terjadi, menjadi tantangan besar bagi ahli anestesi yang belum berpengalaman dalam menangani operasi pemisahan kembar siam. **Tujuan:** Untuk memaparkan manajemen anestesi dan pemanfaatan tele-anestesi pada operasi pemisahan kembar siam. **Laporan Kasus:** Bayi perempuan kembar siam tipe pygopagus, berumur 60 hari dengan berat badan total 7030 gram. Bayi lahir dengan persalinan spontan pervaginam, usia aterm, dengan berat lahir total 4.000 gram. Kedua bayi tampak sehat, bergerak aktif, tidak ditemukan gangguan fungsi respirasi, hemodinamik stabil, dan tanpa kelainan kongenital penyerta. Pemeriksaan *computerized tomography-scan* (CT-scan) daerah sacral menunjukkan bayi kembar siam dengan penyatuan kutis dan subkutis di regio perianal serta tidak tampak penyatuan organ internal-vertebral-spinal. Manajemen anestesi dilakukan oleh dua tim anestesi. Setelah memastikan tidak terdapat sirkulasi silang dengan uji sulfas atropine, induksi anestesi dilakukan secara bergantian dengan teknik inhalasi dengan mempertahankan pernapasan spontan. Pemeliharaan anestesi dengan sevoflurane 2,0 – 3,0 vol%, dengan flow 4 L/menit dalam campuran oksigen dan udara menggunakan Jackson Reese. Volume sirkulasi, stabilitas hemodinamik, dan normotermi dipertahankan selama intraoperasi. Operasi pemisahan berlangsung selama 20 menit, dan total waktu pembedahan pada masing-masing bayi selama dua jam. Ekstubasi sadar segera dilakukan setelah

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pembedahan selesai. Pascaoperasi, pasien menjalani perawatan di PICU dan dipulangkan pada hari ke 11. Selama persiapan preoperative hingga tindakan pembedahan, tim anestesi setempat melakukan konsultasi ke tim anestesi pediatri RSUD dr Soetomo secara telemedicine dan telementoring intra-anestesi. **Kesimpulan:** Persiapan dan evaluasi praoperasi yang cermat, pemeliharaan intraanestesi yang tepat, serta komunikasi dan kerjasama tim yang baik merupakan kunci sukses manajemen anestesi pada operasi pemisahan bayi kembar siam. Konsultasi dan pendampingan secara tele-anesthesia memberikan manfaat yang besar bagi tim anestesi yang belum berpengalaman dalam operasi pemisahan bayi kembar siam.

Kata kunci: Kembar Siam; Komplikasi Bayi Baru Lahir; *Pygopagus; Telemedicine; Tele-anesthesia*

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INTRODUCTION

Conjoined twin is a rare congenital disorder in humans and becomes one of the most difficult to manage. This disorder requires medical and psychological attention and is an example of medicine's most complex problem-organizing and ethical issues (1). The incidence of conjoined twins is 1.47: 100,000 live births (2). Based on the fusion, both dorsal and ventral, conjoined twins can be divided into eight subtypes: omphalopagus, thoracopagus, cephalopagus, ischiopagus, parapagus, craniopagus, rachipagus and pygopagus. Thoracopagus is the most common type (40%), followed by omphalopagus (32%), pygopagus (19%), ischiopagus (6%), and craniopagus (2%) (3).

The management of conjoined twins requires good teamwork by involving various multidisciplinary. Well-organized planning and management, both in anesthesia and surgery, are crucial to obtain a good outcome. Besides the medical aspect, careful ethical and moral considerations involving families, psychologists, religious leaders, and legal experts must also be considered. Publication of the management of conjoined twins to the media also needs to be communicated carefully. Therefore media coverage becomes a constructive potential to increase understanding of conjoined twins in society, mobilize community roles, and increase team spirit instead of becoming a burden (1).

Anesthetic management for conjoined twin separation surgery is an enormous challenge for pediatric anesthesiologists. It should be emphasized that each conjoined twin should be treated as a separate entity with specific problems from each baby; thus, it requires two anesthesia teams for the separation surgery (1,2).

In a health facility performing conjoined twin separation surgery for the first time, an experienced pediatric anesthesia team or a local team with direct assistance from an expert team should handle the anesthetic management. The absence of an experienced pediatric anesthesia team poses an extra challenge for an anesthesiologist without experience in conjoined twin separation surgery. Consultation on the anesthetic management of conjoined twins and intra-anesthesia assistance of an experienced team using telemedicine can solve this problem.

Here, we describe the success of anesthetic management in pygopagus conjoined twin separation surgery performed by an anesthesiologist team without experience in separation surgery. The surgery was performed with remote assistance by an expert anesthesiologist using teleanesthesia. Apart from that, this article describes other

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aspects to consider during the management of conjoined twins.

CASE REPORT

We have obtained written consent from both patients' parents to be included in this report. This conjoined twin separation surgery was the first case performed at Ulin hospital, Banjarmasin. When we received the referral of conjoined twins in our hospital, we immediately formed a multidisciplinary team of 52 people with a pediatrician as the chairperson. This conjoined twins' team regularly met to discuss the management of the conjoined twins.

The twin baby girl with Pygopagus conjoined was born by spontaneous delivery, term, with a total birth weight of 4,000 grams (Figure 1). Both babies looked healthy, moved actively, had no respiratory function disorders, were hemodynamically stable, and had no congenital abnormalities. The laboratory test results were within normal ranges (Table 1). At seven days, a computerized tomography scan (CT scan) of the sacral region showed conjoined twins with skin and subcutaneous unification in the perianal region. It did not reveal the internal-vertebral-spinal organ's unification (Figure 2). Echocardiographic examination obtained normal heart function. These babies underwent treatment in the NICU until the separation surgery was performed.

Since the fusion was limited to the skin and subcutaneous tissue, which required simple surgery, the team decided that our hospital surgical team performed the separation surgery of the conjoined twins without direct assistance from an experienced team. In addition, a spike in daily cases of COVID-19 in Indonesia at the time of the surgery resulted in travel restrictions that made it impossible for an expert team to attend. However, considering the complexity of anesthetic management of conjoined twin separation surgery, we conducted telemedicine consultation and intra-anesthesia remote assistance with the pediatric anesthesiologist team from Dr. Soetomo General Academic Hospital, Surabaya, which has experience managing conjoined twins.

Figure 1. Pygopagus Conjoined Twins. There was Unification in The Pelvic Area. N1 Babies Had A Larger Body Size Than N2 Babies

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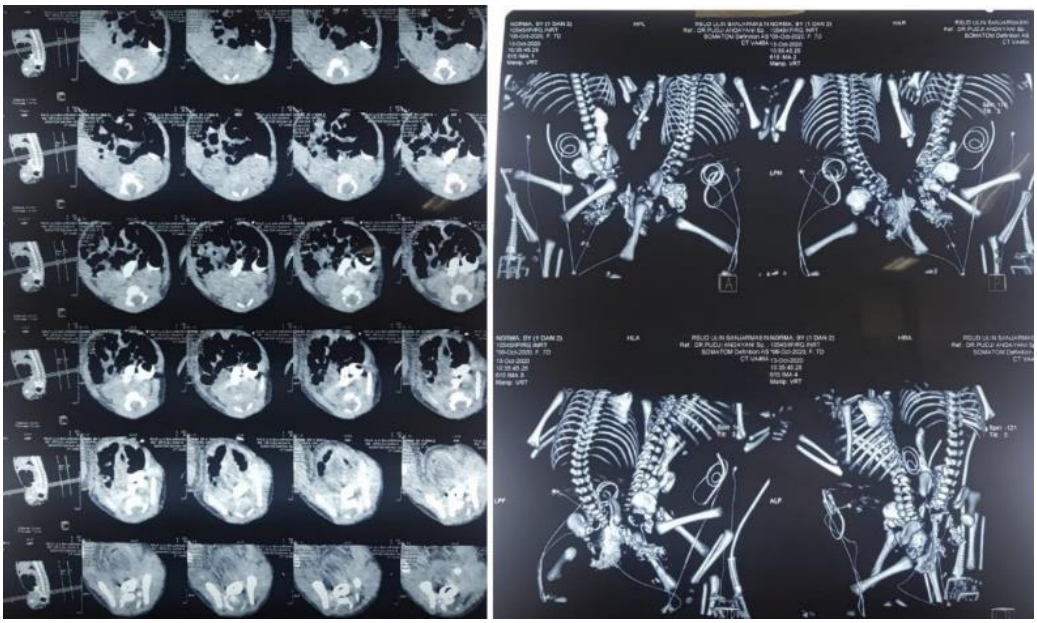

Figure 2. Computerized Tomography-scan (CT-scan) of the sacral region at 7 days of age. It showed conjoined twins with skin and subcutaneous fusion in the perianal area and no visible internal-vertebral-spinal fusion.

Table 1. The Summary of Laboratory Findings.

Laboratory parameters	N1			N2		
	Day 2	Preop	Postop	Day 2	Preop	Postop
Hemoglobin (g/dL)	14.1	15.8	14.5	13.8	15.8	14.6
Hematocrit (%)	42.2	45.7	42.4	39.3	46.3	42.3
White blood count (/µL)	15,300	8,700	7,900	14,100	10,500	7,400
Platelet count (/µL)	271,000	368,000	383,000	336,000	388,000	366.000
SGOT (U/L)	48	24	32	49	27	35
SGPT (U/L)	19	27	28	24	13	26
Ureum (mg/dL)	25	30	29	28	37	25
Creatinin (mg/dL)	0.45	0.18	0.25	0.52	0.24	0.21
Blood glucose (mg/dL)	132	126	123	73	132	122
Albumin (g/dL)		3.9	4.0		4.0	3.7
PT/APTT (second)		10.3/34.6	13.1/39.2		10.9/34.6	11.8/34.3
Na/K/Cl (Meq/L)		137/5.3/113	138/4.6/109		137/5.9/114	138/4.9/110

Before surgery day, we performed two consecutive days of surgery simulations in operating rooms using dolls and involved the entire team in charge (Figure 3). The surgery was performed in two adjacent operating. The first operating room was equipped with two anesthesia machines, two patient monitors, and other anesthesia equipment was doubled. Two anesthesia teams, each comprising one anesthesiologist consultant, carried anesthetic management out.

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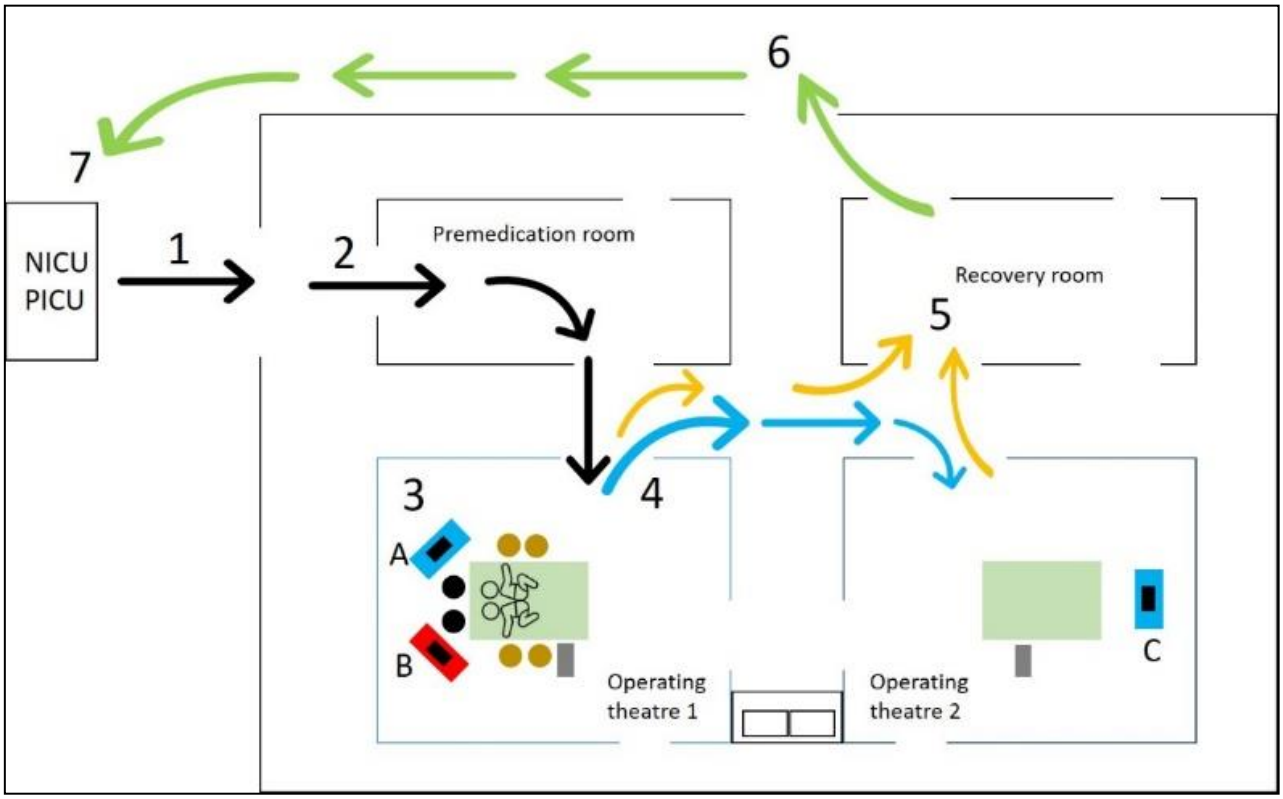

Figure 3. Simulations of A Conjoined twin's Separation Surgery in The Operating Room. The order of procedure, division of tasks, and roles of each team have been determined. Team 1 (pediatricians and infant nurses) is assigned to transfer babies to the operating room, then received by team 2 (anesthesia resident and nurses). The babies are then transferred to the premedication room and operating room 1 (black arrow). The two anesthetic teams responsible for each baby were ready to receive conjoined twins (3). After a successful separation surgery, team 4 (anesthesia resident, nurses, and surgical resident) will transfer N2 babies into the second operating room (blue arrow). The second surgical and anesthesia team has been prepared to receive N2 babies. After the surgery is completed, both babies are transferred to the recovery room by the anesthesia team (yellow arrows) and received by team 5 (anesthesiologist and nurses). Team 6 (pediatrician and infant nurse), together with the anesthesia team of each baby, make a transfer from the recovery room to the PICU (green arrow) and will be received by Team 7 (pediatrician, pediatric resident, infant nurse) in the PICU.
Notes: A; Anesthesia machine, patient monitor, and workstation for infants N2. B; Anesthesia machine, patient monitor, and workstation for babies N1; Anesthesia machine, patient monitor, and workstation for infant N2 in operating room two after separation. Black circle; anesthesiologist position. Brown circle; the position of the surgical team during surgery. Gray square; surgical instrumentation table.

We performed the separation surgery when the baby was 60 days old, with a total body weight of 7030 grams. To avoid confusion, we named the two babies N1 (the bigger baby) and N2. As per the elective surgery protocol in our hospital during the COVID-19 pandemic, two days before surgery, both babies underwent RT-PCR tests, and the results were negative for SARS-CoV-2 for both babies. Both babies had peripheral venous access using a large-bore cannula, and a peripherally inserted central catheter (PICC) was placed in the brachial vein. On arrival at the operating room, routine non-invasive monitors (blood pressure, ECG, peripheral SpO2, temperature) were placed in both babies. Before induction, N1 babies were given 0.03 mg of atropine intravenously. Our observation found no cross-circulation symptoms after atropine administration.

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Induction of anesthesia by incremental administration of sevoflurane was performed on N1. Four µg of fentanyl and 3 mg of propofol were given intravenously as an adjunct to facilitate intubation. We performed successful intubation using a 3.0 micro-cuffed endotracheal tube (ETT) in a lateral position. We performed the same procedure on N2. After intubation, intra-arterial blood pressure (IABP) was placed in the femoral arterial of both babies under ultrasound guidance. Anesthesia was maintained using sevoflurane 2.0-3.0 vol%, with FiO_2 40% in oxygen and air mixture with a total flow of 4 L/min using Jackson Reese.

Infiltration of 1% lidocaine with epinephrine 1:200,000 was administered to the incision site before surgery. The second surgical and anesthesia team was already in the second operating room when the separation was almost completed. The separation surgery lasted for 20 minutes then we transferred the N2 to operating room 2. The total surgical time for both babies was 2 hours with minimal intraoperative bleeding. We immediately performed awake extubation when the surgery was completed. Both babies were transferred to the PICU after ensuring that the hemodynamics were stable and there were no anesthesia residuals in the recovery room. Administration of intravenous paracetamol 50 mg every 6 hours as postoperative analgesia resulted in pain scores of 3 on the FLACC scale in both babies. We found neither baby with surgical wound infection, nerves disorders, limb weakness, or autonomic disorders during hospitalization. Both babies were discharged on day 11 with negative SARS-CoV-2 results.

DISCUSSION

The incidence of pygopagus conjoined twins is around 19% (4) and is more common in females (5). The separation surgery of conjoined twins poses challenges for both surgeons and anesthesiologists (6). The separation surgery should not be performed during the neonatal period to facilitate safety, except in urgency or emergencies (1). Surgery performed during the neonatal period can increase mortality by 50% (7). The separation surgery at infancy can increase the chances of success. However, prolonged procrastination increases the risk of both physiological and psychological problems and increases complications and costs (1).

Peri-anesthesia management in conjoined twin separation includes pre-operative preparation, intra-, and postoperative management. Pre-operative preparation comprises the preparation for the baby and anesthesia equipment and task division for the anesthesia team in the operating room. Preparation for the baby can be divided into several stages. At birth, the resuscitation phase is necessary to stabilize the baby's condition during transportation to the referral center. The next stage is stabilization and diagnostics. Stabilization steps are needed to optimize the baby's condition until the separation surgery is performed (1). Exposure to nosocomial infections during the stabilization stage should be avoided. In our case, to prevent the risk of exposure to nosocomial infections, including being infected with SARS-CoV-2, the conjoined twins were hospitalized in a particular room with a separate nurse who was not in charge of treating other babies. Visits were limited to the baby's parents while maintaining health protocols.

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Diagnostic stages are needed to determine the unification site, unified organ system, concomitant complications, and airway assessment (8). Imaging studies can accurately determine occurring organ division and vascular anomalies. This imaging is essential in determining the surgical plan and prognostic information (5). On pygopagus conjoined twins, an abdominal and pelvic CT scan is required to assess whether organ systems are union. In our case, the CT scan did not show internal-vertebral-spinal organ fusion and shared-circulation.

Preparation of anesthesia equipment such as anesthesia machine, breathing circuit, laryngoscopes, monitors, infusion pumps, syringe pumps, fluid warmer, and temperature control device should be available for each baby (2,9). The anesthesia team's position and machine placement are discussed with the surgical team. The placement of all equipment in the operating room must be designed according to the type of conjoined twins. In pygopagus conjoined twins, the anesthesia machine is placed on the same side of the head of the operating table (9). All anesthesia equipment for each baby is marked with two different colors, likewise with the drugs used.

The presence of cross-circulation and difficulty in managing the airway should be considered when performing anesthesia induction (9,10). In our case, although the CT scan revealed no fusion of the organ systems, we performed an atropine test to confirm that there was no cross-circulation (1). Difficult intubation was commonly found in the thoracopagus type due to the position of the babies facing each other (11). In the pygopagus type, difficult intubation can occur due to the lateral position of the baby. After ensuring there was no cross circulation, we alternately performed anesthesia induction and intubation on both babies. In anticipating airway management difficulties, we intubated the baby under deep sedation using sevoflurane inhalation and intravenous adjuvant while maintaining spontaneous breathing. Anesthesia induction in conjoined twins can be performed both by inhalation and intravenously. It can be done by considering the presence or absence of intravenous access before induction, predicting difficulty in airway management, and the anesthesiologist's preference (9). Muscle relaxants can be administered after the airways of both babies have been secured (2). Intraoperative hemodynamic stability and circulating volume should be maintained. Determining the amount of blood loss from each baby is quite tricky, especially in conjoined twins with cross circulation (1,10). Fluid replacement and blood loss can be based on visible blood loss through a suction tube, weighed gauze, assessment of peripheral perfusion and urine production, serial hematocrit, measurement of intra-arterial blood pressure, and central venous pressure (1,9,10). Hypothermia should be avoided because of prolonged surgical duration, operating room temperature, and heat loss through surgical wounds (7,9). We covered both babies' bodies using a heating mattress and intravenous fluid warmers to prevent hypothermia.

Massive blood loss and replacement, long-duration surgical procedures, and pre-operative anatomical changes can cause postoperative problems (2,9,10). Long-duration surgical procedures, unstable hemodynamic conditions, and continuous blood loss in conjoined twins of thoracopagus type will require postoperative mechanical ventilation (2). In our case, because the

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unification was only in the skin and subcutaneous tissue and underwent a simple surgical procedure, we performed extubation as soon as the surgery was complete. Postoperative care was carried out in the PICU while maintaining hemodynamic stability, fluid and electrolyte balance, and preventing infection.

Anesthetic management in a conjoined twin is different from that of normal infants. In our case, although the fusion is only of skin and subcutaneous tissue and will undergo a simple surgical procedure, this is our first case regarding the separation of conjoined twins. Direct assistance by an experienced pediatric anesthesiologist team will be required. Because direct assistance by an experienced pediatric anesthetist team was not possible, we conducted consultations and discussions to manage conjoined twins with the pediatric anesthesia team of Dr. Soetomo Hospital with telemedicine since receiving a referral for conjoined twins to postoperative care. We conducted this telemedicine consultation using the WhatsApp messaging facility and the Zoom video conferencing (Zoom Version 5.0.2; Zoom Video Communications Inc., San Jose, CA, USA). Likewise, during surgery, intra-anesthesia assistance was carried out by teleanesthesia using the facilities of the WhatsApp messenger application. Dr. Soetomo's pediatric anesthesia team has been experienced in managing conjoined twins since 1975. Until 2017, this team has handled 85 cases of conjoined twins (1).

During the COVID-19 pandemic, remote consultation using communication technology has increased (12). Using an online communication system is also beneficial for continuing the anesthesia trainer education program during the pandemic (13). Telemedicine has been known since the 1970s.

The WHO defines telemedicine as the provision of health care services through communication technology for disease diagnosis and treatment and for continuing education of health care providers where distance is a constraining factor (14).

Teleanesthesia, which is the use of telemedicine in anesthesia, has grown in the last few decades. This development is inseparable from the support of advances in communication technology. Teleanesthesia was first reported in 2004 (14) as a pre-operative evaluation consultation on ten patients in a remote facility, carried out by a nurse using a camera (15). In the same year, an anesthesiologist based in Virginia reported an intraoperative telemonitoring on patients undergoing laparoscopic cholecystectomy surgery in Ecuador (16). In 2009, Fiadjoe et al. (17) reported the successful use of teleanesthesia in 2 cases of liver transplant surgery. The Philadelphia-based anesthesia team mentors the team in India. Using teleanesthesia provides benefits such as reducing travel costs, facilitating specific consultations for diagnosis, therapy, and prognosis, facilitating postoperative monitoring, ICU care and rehabilitation, and educational and training purposes (18).

Other aspects to be considered during managing conjoined twins are communication and teamwork, ethical and legal aspects, and media coverage. The management of conjoined twins involves many multidisciplinary team members. Good communication and cooperation are the keys to successfully managing conjoined twins (1,8). To obtain good coordination and division of tasks during the separation surgery,

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we performed two simulated surgeries with a doll and performed in the operating room before the surgery day. Each team performed its respective duties and roles (1).

Conjoined twins have always attracted the attention of both the public and the medical community. Excessive media coverage can negatively affect the patient's parents and burden the medical team (1). In our case, starting from the referral process to postoperative care, we did not publish the management of conjoined twins in the mass media out of respect for the rights of the patient's family. We published to the media the successful management of conjoined twin separation when both babies were discharged from the hospital (19).

CONCLUSION

Conjoined twin separation surgery presents challenges for pediatric anesthesiologists. Although fusion is limited to the skin and subcutis, the possibility of cross-circulation and the prediction of difficult intubation requires careful attention and monitoring. Teleanesthesia as consultation and assistance means from an experienced anesthesiologist team provides significant benefits for the anesthesiologist without experience in conjoined twin separation surgery. Good communication and teamwork are essential to successful management in conjoined twin separation surgery. Ethical and legal aspects involving parents and families and the right communication media can potentially construct and increase team spirit.

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