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Case Report

Anesthetic management of pygopagus conjoined twins: techniques to evaluate cross-circulation

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Abstract: Given the associated intricacies associated with surgery and anesthetic management, the care of conjoined twins requires a multidisciplinary approach. Due to the complexity of these patients, anesthetic care may be required for radiologic imaging, placement of tissue expanders, insertion of permanent or temporary vascular access devices, and definitive separation surgery. We present a case of pygopagus conjoined twins with multiple anesthetic encounters, including definitive separation surgery. The perioperative management of such patients, with emphasis on the shared circulation and airway management, is discussed.

Keywords: Pediatric anesthesia, conjoined twins, pygopagus

Introduction

More than 60% of conjoined twins succumb in utero or are stillborn, resulting in an estimated prevalence of approximately 1:250,000 live births. However, the actual incidence of conjoined twins may be decreasing, given early diagnosis and the option for termination of pregnancy [1]. Conjoined twins are monozygotic, mono-amniotic, and mono-chorionic, and are always of the same gender, with females outnumbering males by a ratio of 3:1 [2]. Controversy still remains as to whether their formation results from failure of separation or subsequent fusion [3, 4]. Conjoined twins are always joined at homologous sites and are classified by the site of attachment.

Long term survival and the potential for surgical separation depend on the attachment site, degree of major organ and circulation sharing, and the presence of other congenital anomalies. Therefore, adequate evaluation to identify the extent of shared organs and systems is essential. During the evaluation process, the twins may require sedation or general anesthesia several times for imaging studies, vascular access placement, and other preparatory studies or surgical procedures prior to definitive separation. We present a case of pygopagus conjoined twins who required anesthesia or sedation nine times prior to separation. The perioperative management of such patients, with emphasis on the shared circulation and airway management, is discussed.

Case report

Institutional Review Board approval for single case reports is not required by Nationwide Children's Hospital (Columbus, Ohio). A pair of 3-month-old, female, 11.2 kg (combined weight) conjoined twins fused at the posterior pelvis (pygopagus) was transferred from Uganda to our institution for evaluation for separation surgery. Previous surgical history included colostomy placement in Uganda on day 5 of life in each twin due to imperforate anuses. On physical examination, both twins were well-developed and had a normal appearance with

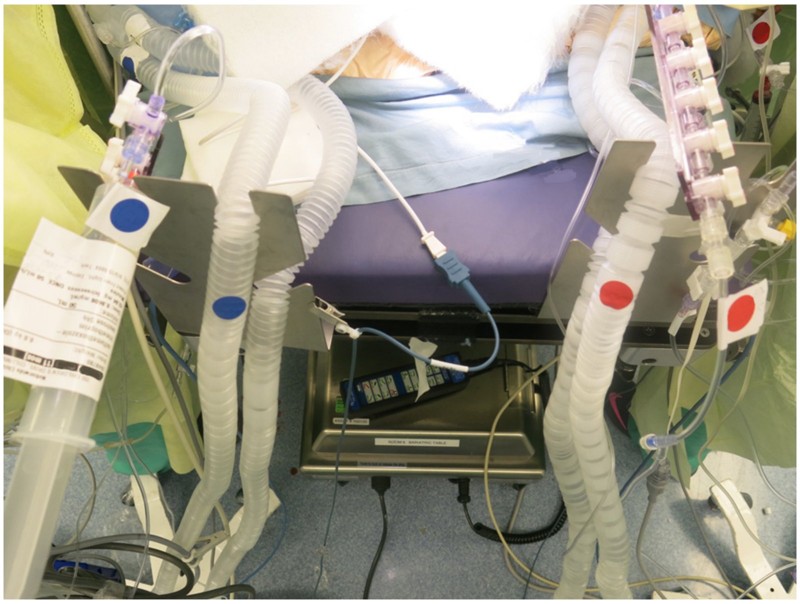

Figure 1. Photograph showing the color coding of the circuits and all supplies.

the exception of being conjoined. Each of the infants had separate vaginas and urethras, but no anal openings. Echocardiogram did not reveal associated congenital heart disease.

The initial studies performed included magnetic resonance imaging (MRI), pelvic examination, cystoscopy, and vaginoscopy. For the initial procedure, two anesthetic machines were prepared in a single operating room (OR) equipped with two separate anesthesia booms with adequate gas outlets. Two anesthesia teams with two different anesthesia work-spaces and monitoring equipment were used. All anesthesia equipment, including medications, respiratory circuits, infusion lines, and anesthesia providers were color coded and labeled as blue for twin A and red for twin B, as were the twins themselves (**Figure 1**). A 24 gauge intravenous (IV) cannula was placed for each twin prior to anesthetic induction. The twins were transported to the operating room, and standard American Society of Anesthesiologists' (ASA) monitors were placed for each. For the purpose of medication administration, the body weight for each was calculated as one-half of the combined weight. Following pre-oxygenation with 100% oxygen to both infants, anesthesia was simultaneously induced with propofol for twin A and propofol plus fentanyl for twin-B, followed by bag-valve-mask ventilation with sevoflurane in oxygen. The twins were rotated slightly laterally so that twin A's trachea could be intubated with an oral 3.0 mm cuffed endotracheal tube (cETT). While twin A's trachea was intubated, bag-valve-mask ventilation was provided for twin B. After endotracheal intubation of twin A, the trachea of twin B was intubated with an oral 3.0 mm cETT. To test the degree of cross-circulation, atropine (0.035 mg/kg) was administered to Twin B after endotracheal intubation. Although twin B's heart rate (HR) increased from 150 to 170-180 beats/minute (bpm) soon after the administration of atropine, no change was noted in the HR of twin A, which remained at 160 bpm. After anesthetic induction, the twins were transported to the MRI room in the OR where two MRI-compatible anesthetic machines had been prepared. Anesthesia during MRI was maintained with sevoflurane (end-tidal concentration of 3% in 50% oxygen in air). After the 2.5 hour MRI, the twins were transported to the operating room where cystoscopy and vaginoscopy were performed. To further assess the amount of shared or crossed circulation during those procedures, 50% nitrous oxide in oxygen was administered to Twin A (total fresh gas flow of 2 liters per minute). No exhaled nitrous oxide was noted from Twin B during the 30-60 minute procedure. Glycopyrrolate (10 μg/kg) was also administered to Twin-B to detect cross-circulation. While the HR increased in Twin B, no hemodynamic change was observed in Twin A. At the completion of the procedures, the tracheas of the twins were extubated, and they were transferred to the post anesthesia care unit (PACU).

The second surgery, insertion of tissue expanders, was performed 3 weeks later. The twins were transported to the OR with peripheral IV cannulas in place. Anesthesia was simultaneously induced for both twins through separate IV cannulas with propofol and fentanyl followed by neuromuscular blockade with rocuronium. The patients' tracheas were intubated sequen-

Figure 2. Photograph showing the anesthetic circuits as they are crossed and attached to the patients (top of the photograph) during prone positioning.

tially as had been performed for the first surgery. After a full body, circumferential sterile prep was performed; the twins were placed in the prone position. During prone positioning, IV infusions were moved to each patient's side while the respiratory circuits were crossed (**Figure 2**). While the twins were in the prone position, the anesthesia machine for twin A was on the twin B side, and a medication error occurred. An anesthesiologist assessed one the twins that was anesthetized and determined that additional analgesia was needed. Despite the color-coding of the IV infusion lines, morphine was administered to the wrong twin. As this occurred early in the procedure, there were no sequelae. Two tissue expanders were placed posteriorly. Again, the separation of their circulation was demonstrated, as one twin showed vital sign changes including tachycardia and increased blood pressure in response to epinephrine in the local anesthetic agent, which was injected into the skin and subcutaneous tissue at the site of insertion of the tissue expander, while no changes were noted in the other twin. The twins were then turned back to the supine position, and an additional tissue expander was placed anteriorly. After tracheal extubation in the OR, they were transported to the PACU. Their postoperative course was unremarkable.

Subsequently, there were 6 additional procedures prior to the final separation (**Table 1**). Although most of those procedures were performed under general inhalational anesthesia, revision of a percutaneous intravenous central catheter (PICC) line was performed under general anesthesia for one twin with sedation for the other twin whose PICC line did not require revision. For twin A, who required PICC insertion, anesthesia was induced with propofol (3.5 mg/kg) and a laryngeal mask was inserted followed by maintenance anesthesia with sevoflurane (end-tidal concentration 3%). Although no procedure was performed for twin B, sedation was required to provide immobility to allow the procedure for twin A. As such, twin B was sedated with a single bolus of dexmedetomidine (0.5 μg/kg) after anesthesia had been inducted for twin A. During anesthetic induction for twin A, spontaneous ventilation was maintained with supplemental oxygen administered via a nasal cannula for twin B further demonstrating the relative separation of their circulations.

Separation surgery was performed at 11 months of age when the twins weighed a combined 17 kilograms. Prior to the surgery, diag-

Table 1. Summary of surgical and anesthetic history of the patients

Surgery	*Age at procedure*	*Height and weight**	*Position*	*Estimated blood loss*	*Procedure and (anesthesia) time*	*Anesthesia notes*
Colostomy	5 days	Not available	Supine	Minimal	Not available	N/A
MRI, pelvic EUA, cystoscopy, and vaginoscopy	3 months	55 cm/11.2 kg	Supine	Minimal	2:44 (4:21)	HR response from atropine following injection into peripheral IV was detected only in twin A. Nitrous oxide was administered to one twin and no exhaled nitrous oxide was noted from second twin.
Insertion of tissue expanders	3 months	55 cm/11.6 kg	Supine and prone	Minimal	3:01 (5:28)	Increased HR following the subcutaneous injection of epinephrine in the local anesthetic solution was noted only in one twin. Medication error occurred after positional change.
I&D of tissue expander with wash out due to local infection. PICC insertions.	4 months	55 cm/11.8 kg	Supine and prone	Minimal	1:10 (3:02)	-
Removal of infected tissue expander	4 months	55 cm/12.4 kg	Prone	Minimal	0:35 (1:23)	-
I&D of back wound	5 months	62 cm/14.0 kg	Prone	Minimal	1:13 (2:06)	-
Insertion of tissue expander to the posterior trunk	6 months	64 cm/14.6 kg	Prone	Minimal	1:45 (2:29)	-
PICC insertions	8 months	66 cm/16.0 kg	Supine	Minimal	0:25 (0:54)	-
PICC insertion	9 months	67 cm/16.8 kg	Supine	Minimal	0:15 (0:58)	End-tidal sevoflurane of 3% and propofol (3 mg/kg) were administered to twin A with no response in twin B. Twin B was awake and required sedation with dexmedetomidine while general anesthesia was provided for twin A.
Separation surgery (Removal of tissue expander, separation of spinal cord, anal pull through, and perineal/buttock reconstruction)	11 months	70 cm/17 kg	Prone	200 mL for each twin	10:55 (15:59)	Anesthesia was induced and neuromuscular blockade provided for twin B while spontaneous ventilation was maintained in twin A with no change in the BIS value.

*The weight listed is the combined weight at the time of procedure. For medication dosing for each twin, the weight was divided by 2. MRI = magnetic resonance imaging; EUA = exam under Anesthesia; I&D = incision and drainage; PICC = percutaneous intravenous central catheter; NA = not available; HR = heart rate; IV = intravenous; BIS = bispectral index.

Table 2. Pharmacologic demonstration of cross-circulation

1. Atropine and glycopyrrolate: Heart rate response in one twin and no response in the other.
2. Nitrous oxide: Nitrous oxide administered to one twin and no exhaled nitrous oxide was noted from second twin.
3. Sevoflurane: End-tidal sevoflurane of 3% was administered to one twin with no response in the other twin. The second twin was awake and required sedation with dexmedetomidine while general anesthesia was provided with sevoflurane for PICC placement in the other twin.
4. Propofol: Intravenous induction of anesthesia with propofol resulted in no change in level of consciousness in other twin. Additionally, during the separation surgery, there were discordant values on the bispectral index (BIS) of the two twins when anesthesia was induced in twin B while twin A remained awake with no change in the BIS.
5. Epinephrine: During placement of tissue expanders, hemostasis and cutaneous analgesia was provided by the administration of a local anesthetic solution with epinephrine. Hemodynamic changes (increases in heart rate and blood pressure) were noted in one twin with no changes in the other.
6. Rocuronium: During the definitive separation surgery, anesthesia was induced and neuromuscular blockade provided for twin B while spontaneous ventilation was maintained in twin A until after the airway was secured via nasotracheal intubation in twin A.
7. Calcium chloride: During the definitive separation surgery, several doses of calcium chloride were required in the one twin while no change was noted in the plasma ionized calcium concentration of the other twin.

Figure 3. Induction technique of individual infants at the time of separation surgery. Given the relative separation of their circulations, there was limited impact on twin A of the medications used for anesthetic induction on twin B. No change was not in the bispectral index of twin A despite the induction of general anesthesia with propofol and an end-tidal sevoflurane concentration of 3% in twin B.

nostic imaging demonstrated that they had fusion of the lower portion of their spinal cords. Prior to the separation surgery, meetings and simulations were held with all team members present. Two separate anesthesia teams with duplication of all anesthetic equipment and medications were prepared and color-coded in the OR. The twins came to the OR with PICC lines in place. Standard ASA monitors and bispectral index (BIS) monitors were applied for each twin. The baseline BIS was 75-80 in both twins. Hydromorphone (20 μg/kg) was administered to twin A for sedation. Anesthesia was induced in twin B with propofol (3 mg/kg) and fentanyl (1.5 μg/kg) followed by neuromuscular blockade with rocuronium (0.6 mg/kg). During bag-valve-mask ventilation and nasotracheal intubation (3.5 mm cETT) with standard direct laryngoscopy for twin B, twin A was breathing spontaneously with supplemental oxygen via an anesthesia mask and circuit (**Figure 3**). Although the BIS decreased to 40-50 for twin B, the BIS for twin A remained at the baseline value of 75-80. After the ETT was secured for twin B, anesthesia was induced in twin A with propofol (4 mg/kg) and hydromorphone (30 μg/kg) followed byneuromuscular blockade with rocuronium and nasotracheal intubation. A radial arterial cannula and additional peripheral intravenous access was secured for each twin in their upper extremities. After a full body circumferential prep was performed, Foley catheters were placed, and neurophysiological monitoring for spinal cord integrity was achieved. As the twins were turned prone for the start of the surgical procedure, the anesthesia circuits were disconnected and switched so that all anesthesia equipment including the anesthesia machines, respiratory circuits, monitoring leads, and venous/arterial cannulas were on the correct side (with color coding, as before). Maintenance anesthesia included isoflurane (end-tidal concentration of 0.6-1.5% in 50-60% oxygen in air) with incremental doses of hydromorphone as needed. The surgical team consisted of pediatric surgeons, plastic surgeons, and neurosurgeons. The surgical team completed separation of the conjoined twins after 8 hours and 45 minutes. Twin B was then moved to a separate OR table, after which the surgical, anesthesia, and nursing teams separated into two groups on opposite sides of the same OR. Surgical reconstruction and primary wound closures were then performed for each twin. Total operative time was 16 hours. Intraoperative fluids and blood products included 568 mL and 350 mL of packed RBCs, 750 mL and 700 mL of colloid, and 1700 mL and 1550 mL of crystalloid for twin A and twin B, respectively. A single dose of 10 mg/kg of calcium chloride was administered to twin B due to a low ionized calcium level. The ionized calcium level of twin A remained in the normal range throughout the procedure. No vasopressor or catecholamine support was required during the procedure. Body temperatures measured at the mid-esophagus prior to separation and at the end of surgery were normal (37.1°C and 36.9°C) and (36.7°C and 36.1°C) at each time for twin A and twin B respectively. The separated twins were transported to the ICU with the endotracheal tubes in place and mechanical ventilation was provided postoperatively. Twin A and twin B had tracheal extubation on postoperative days (POD) 3 and 2, respectively. Postoperative analgesia was achieved with fentanyl, administered by nurse-controlled analgesia (NCA). Twin B required two subsequent surgical procedures on POD 7 and 16 for management of a cerebrospinal fluid leak. The remainder of their postoperative course was uncomplicated.

Discussion

Pygopagus attachment occurs in approximately 19% of all conjoined twins [2]. Since pygopagus twins are fused at the sacrum, the buttocks, and the perineum, there are generally fewer concerns regarding the potential fusions of vital organs such as the heart or liver. However, pygopagus twins can demonstrate communication between their sacral venous plexus resulting in a shared circulation. As noted in our patients, issues with airway management during anesthetic induction are relatively minimal compared with other types of conjoined twins. In our patients, we were able to rotate them laterally to allow for easy bag-valve-mask ventilation of one twin while tracheal intubation was performed for the other. Eventually, as we determined that their circulations were relatively separate, we were able to induce anesthesia in one with maintenance of spontaneous ventilation in the other. Given the limited sharing of major organ systems with pygopagus twins, separation is generally more feasible and the survival rate is higher than with other types of conjoined twins [2]. However, a careful and thorough preoperative evaluation is mandatory to evaluate for shared organ systems, generally involving the vertebral bodies, spinal cord, or anorectal region as was noted in our patient [5]. When caring for such patients, anesthesia care may be required for imaging studies, placement of chronic indwelling vascular access lines, and for the definitive separation surgery. Additionally, in cases with associated rectal anomalies, colostomy may be required in the immediate neonatal period. The anesthetic and surgical management of conjoined twins has been previously described by various centers [7-12]. During the preoperative assessment and anesthesia planning, factors to be considered include the site of attachment, the sharing of vital organs and circulation, as well as other associated anomalies. All of these factors can impact anesthetic management regarding the choice of anesthetic agents, induction technique, airway management, monitoring, vascular access and positioning not only of the twins, but also of the anesthesia, surgical, and nursing teams.

Planning and simulation

Surgery for the separation of conjoined twins is both exceedingly rare and extremely complicated, requiring the cooperation of a large number of medical, nursing, and allied personnel. As such, prospective planning is vital, especially for the definitive separation surgery. Good communication between the staff remains an integral component of ensuring success in such endeavors. We believe that there is a need to identify a lead surgeon who acts to coordinate all activity of the team. In planning meetings, the following should be discussed:

1. Results of all laboratory and radiologic investigations. 2. The order in which different surgical specialties will operate. 3. Planned changes in patient positioning. 4. Location of intravenous and arterial cannula as well as monitoring devices. 5. Anticipated problems or concerns. 6. Postoperative management.

After the meetings, a full environmental simulation is suggested to optimize anesthesia and surgical flow, optimize positioning of all teams in the operating room, ensure that the needed equipment and support is present in the OR to be used, identify unanticipated problems, promote leadership and communication, and ensure safe, effective patient care without complications or delays [13].

Separation of teams and twins using color-coding

Each twin should be considered as a separate patient. As such, all equipment needs to be duplicated including anesthesia machines, outlet sources of oxygen, nitrous oxide, medical air, anesthesia monitors, and intravenous cannulas. Although the anesthesia medication cart can be shared to save space, we prefer to use two different carts and set-ups. Duplication in the operating room should also include the anesthetic team and the electronic medical record, so that record keeping is separate for each patient. In our case, this required coordination of care with our operating room technology support team to allow for the use of a separate electronic medical record device for each twin. Color-coding of each twin, all equipment, and medications can be used to keep the two teams separated. Despite such precautions, as demonstrated by our experience, mistakes can still happen when two patients are in close proximity to each other. Color-coding for each anesthesia and surgery team is also useful when the twins are eventually separated. Although separate anesthesia machines and ventilators are generally used, a unique publication reported simultaneous ventilation of conjoined twins using a Carlen's Y-adaptor with a single ventilator [14]. The authors concluded that the system was useful, although they noted that the twins had different values of end-tidal carbon dioxide during MRI due to differences in tidal volumes, compliance, and resistance of the respiratory systems between the twins.

Positioning

Intraoperative positioning requires careful planning, simulation, and consideration. It is vital for all members of the team including surgeons, nurses, and anesthesia providers to ensure that there is ample space for all in a crowded operating room around two small patients. Changes in patient positioning during surgery increase the risk of displacement of endotracheal tubes, dislodgement of vascular cannulas, and medication errors. When feasible, the equipment and anesthesia machines should be kept on the same side as the patient; however, this may not be practical when switching from the supine to the prone position. During the definitive separation surgery, the eventual impact of moving the tables apart should always be kept in mind to facilitate this important aspect of separation.

Airway management

Even in the absence of major airway anomalies, airway management can be challenging due to positioning issues as well as the need to support ventilation of the conjoined twin when there is extensive cross circulation. Although airway issues are limited with pygopagus twins; when the twins are facing each other or their neck position is distorted (thoracopagus and craniopagus twins), airway management can be problematic. Especially with thoracopagus twins, one should expect some degree of neck hyperextension and limited space depending on the relative position of the heads and faces. Airway management strategies such as fiberoptic intubation or unique positioning techniques may be helpful if the airway proves to be challenging [15]. For short procedures, a supraglottic device may provide adequate airway control while alternative techniques may be required for endotracheal intubation in scenarios where direct laryngoscopy is not feasible [16]. Given the potential ability to secure the ETT more effectively, we chose the nasal route for endotracheal intubation during the definitive separation surgery during which positioning changes would be necessary.

Vascular access and blood loss

As repeated procedures are frequently required in these patients, vascular access may be challenging. In recent years, the routine use of PICC lines has increased and facilitated patient care. Alternatively, percutaneous central venous catheters may be placed at the time of surgery although access to sites and positioning may be limited depending on the site of attachment. Ultrasound may be invaluable for peripheral and central venous access as well as placement of arterial cannula. Given the potential for blood loss and the need to transfuse fluids, blood and blood products, adequate vascular access is required for such procedures.

Significant blood loss should be anticipated during separation surgery. The amount depends on the site of connection, the organs shared, and the extent of surgery. Close monitoring of coagulation should be initiated preoperatively and continued throughout the procedure. Blood and blood product administration should be guided by ongoing laboratory assessment of hemoglobin, platelet count, and coagulation profile. Such concerns are particularly relevant when the twins share vessel-rich organs such as the liver, heart or skull. In such cases, there may be extensive cross-circulation, thereby making it difficult to recognize the amount of blood loss from each individual twin. Close monitoring of hematologic and coagulation status with point-of-care testing for each twin is suggested. In our patients, arterial access was achieved for the definitive separation surgery not only to provide ongoing hemodynamic monitoring, but also for vascular access for intermittent laboratory sampling. Rapid, bedside point-of-care testing was used to provide immediate results for arterial blood gases, electrolytes, ionized calcium, and hemoglobin.

Temperature management

Hypothermia can easily result while the patients are uncovered during anesthetic induction and placement of venous and arterial cannulas, patient positioning, and full body prep. The temporary loss of skin cover can be considerable during separation surgery. These factors increase heat loss by evaporation, radiation, and convection. To keep the infants warm, they must be transported appropriately wrapped and cared for in apre-warmed operating room. All of the usual means to maintain normothermia should be used including warming of intravenous fluids and forced air warming devices, with coverage of exposed areas with plastic as allowed by the surgical procedure.

Cross circulation

When providing anesthetic care, one of the primary concerns is the potential for cross-circulation between the twins so that medications administered to one may affect the other. Once separation surgery is planned, evaluation of the extent of the cross circulation is critical. Cross circulation between conjoined twins is much more common among thoracopagus and craniopagus twins. In those cases, angiography or radioisotope imaging for cross circulation is necessary for estimation of each individual twin's cardiac output percentage, as one of the twins might be dependent on the other's circulation for survival [17].

Even with accurate imaging studies, the degree of cross-circulation may be difficult to determine unless large vascular or cardiac connections are noted. While significant cross-circulation can be detected through imaging studies such as MRI, CT or angiography, it can be also detected by injection of radioactive albumin, methylene blue, or indigo carmine. The degree of cross-circulation significantly impacts many aspects of anesthetic care, most importantly the induction technique. If the anesthetic medications and neuromuscular blocking agents administered to one twin will affect the other, it may be necessary to manage both airways simultaneously. This requires bag-valve-mask ventilation of one infant while endotracheal intubation is being performed on the other. In our patients, the lack of significant cross-circulation was determined during the initial anesthetic encounters and confirmed during later procedures as outlined in **Table 2**.

During our first procedure that required anesthetic care, atropine was administered to one twin resulting in the expected chronotropic response, while no heart rate change was noted in the other twin. A similar experience has been previously reported [6]. Although likely more of a theoretical concern, it has been suggested that paradoxical bradycardia may result if a small dose of atropine is administered related to preferential blockade of prejunctional M_2 receptors with an augmentation of vagal tone [18, 19]. As such, with limited cross-circulation, a sub-therapeutic plasma concentration of atropine could result in the conjoined twin with the theoretical risk of bradycardia. In our patients, the lack of cross-circulation was noted following injection of an intravenous agent (Propofol), administration of inhalational agents (sevoflurane and nitrous oxide), and a neuromuscular blocking agent (rocuronium). Despite an end-tidal concentration of sevoflurane of 3% in one twin, the other twin was awake and in fact required dexmedetomidine for sedation. When one twin was receiving nitrous oxide, no end-tidal nitrous oxide was noted in the other twin. The lack of response to Propofol in the conjoined twin was demonstrated not only by the lack of clinical response, but also by the lack of change in the BIS number. The use of the BIS number to evaluate cross-circulation has been reported once previously in thoracopagus twins [14]. During anesthetic induction, sevoflurane was administered to one twin by mask while the other twin received only oxygen. Both were anesthetized as noted by clinical signs as well as changes in the BIS number.

Summary

Although anesthesia for conjoined twins has been reviewed previously, the need to provide such care is still exceedingly rare. For many involved, the care of such patients may be their first experience. Open and thorough discussion, planning, and simulation are integral components to the success of these rare procedures. Given the complexity of such patients, anesthetic care may be required for radiologic imaging, placement of vascular access devices, and for the definitive separation surgery. Keys to care during separation surgery include techniques for airway management, identification of the degree of cross-circulation, obtaining adequate venous and arterial access, maintenance of normothermia, and intraoperative positioning. In the series of anesthetic encounters in our patients, the lack of cross-circulation was detected by varying response to medications including anticholinergic agents, intravenous anesthetic agents, inhalational anesthetic agents, and neuromuscular blocking agents.

Disclosure of conflict of interest

None.

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