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Original Research Article

Conjoined Twins' Separation in West Africa: Experiences from Five Countries

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Abstract: ***Background*:** Conjoined twins (CTs) are rare and Anesthesia may be required for diagnostic imaging, invasive procedures and/or separation. This article described the anesthetics for CTs' separation performed locally in West Africa from 2016 to 2022. Anesthetic management for the separation of conjoined twins performed from 2016 to 2022 in five West Africa countries, Benin, Burkina Faso, Guinea, Niger and Senegal, are reported on. ***Methods*:** Anesthetic management for the separation of conjoined twins performed from 2016 to 2022 in five West Africa countries, Benin, Burkina Faso, Guinea, Niger and Senegal, are reported on. The data collection was done retrospectively. Each country was sent their data to Burkina Faso for study. The parental consents were obtained for surgery and publication. ***Results*:** During the period, nine conjoined were reported. The cases were: 5 omphalopagus, 2 pygopagus, 1 xipho-omphalopagus and 1 thoraco-omphalopagus. Inhalation anesthesia or propofol combined with IV opioids were used. Monitoring relied heavily on pulse-oximetry. Manual ventilation was used in five separations. Separations were achieved in 1.5-4 hours. The death of a twin triggered surgery in one pair and three other babies died after the surgery. The fourteen remaining babies developed well. Peri-operative difficulties revealed widespread difficulties in Africa. ***Conclusion*:** CT separation surgery still constitutes an extraordinary challenge in low and middle income countries, but well-trained anesthesiologists, a multidisciplinary approach and international contacts can lead to safe performance of complex neonatal surgeries.

Keywords: Conjoined Twins, Separation, Pediatric Anesthesia, West Africa.

INTRODUCTION

Conjoined twins (CTs) are rare [1], and Anesthesia may be required for diagnostic imaging, invasive procedures and/or separation. Surgical separation feasibility and approach greatly depends on the organs being shared. Even for the most favorable forms, success of surgery depends on accurate preoperative assessment by imaging techniques and demands good multi-disciplinary collaboration. In West Africa, CTs were usually evacuated to Europe for separation. This article described the anesthetics for CTs' separation performed locally in West Africa from 2016 to 2022.

METHODS

The authors reported their first experiences of CTs' separations performed from 2016 to 2022 in five West Africa countries, Benin, Burkina Faso, Guinea, Niger and Senegal. The data collection was done retrospectively. Each country was sent their data to Burkina Faso for study. It's concerned preoperative, intraoperative and postoperative data. Outcome for each conjoined twins was indicated. Informed written consent was obtained from all parents for being included in the study.

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The authors did not request authorization from the ethics committee for each country because this is not usual for case report in West Africa.

RESULTS

Preoperative Period

Nine CTs were separated: 5 omphalopagus, 2 pygopagus, 1 xipho-omphalopagus and 1 thoraco-omphalopagus. All were transferred to the hospital where they were operated after birth. The clinical examinations were normal except in one baby who presented a respiratory distress. Cross-circulation between babies was estimated in two couples only.

Our multidisciplinary preparatory teams included surgeons, pediatricians, anesthesiologists, radiologists and social workers. The teams met elsewhere then in the OR and no simulation or other novel preparation method was used, however the teams discussed on the bedside how to perform the intubation. The blood banks did not participate in discussions about these surgeries, but they were contacted to make blood available.

Three Separations Were Emergency Surgeries:

One for the neonatal death of a twin, one for ruptured omphalocele and one for a severe infection in one baby. Table 1 details the peri-operative evolution of each pair of CTs.

Intraoperative Period

Electricity, water, oxygen and suction were available in all these hospitals. Each baby had a dedicated anesthesia team leaded by Physician Anesthetist (PA), assisted by Resident Physician Anesthetists (RPA) and/or Non-Physician Anesthetists (NPA) (**Table 1**). Anesthesia equipment was duplicated for each twin also. However, there was no adapted non-invasive blood pressure cuffs during five separations. All the babies benefited from one peripheral intravenous access, none from a central veinous or arterial access. A cross-circulation was revealed in one couple as during the sequential induction in the first baby, the second one became sleepy.

Airway difficulties included one impossible oro-tracheal intubation in one baby in whom a tracheal tear was suspected when the tip of the tracheal tube was palpated under the skin and a gentle ventilation provoked cervical swelling, parents could not afford an ENT exam. Inadvertent extubation while adapting twins' position to the evolving procedure occurred in another one. Mask ventilation was uneventfully maintained until the end of surgery for these two babies. One difficult oro-tracheal intubation was addressed by the using of a rigid chuck. Fluid maintenance used saline, or Ringer solutions added with glucose. After complete separation, the surgery kept going on separated tables in the same room. Major intraoperative complications were significant hemorrhage needing transfusion in one baby, and a bradycardia soon followed by cardiac arrest successfully reversed with CPR and intra-tracheal and IV adrenalin, hypotension (blood pressure of 41/18 and 46/36 mmHg in two babies) addressed by ephedrine and fluid administrations. All babies awoke rapidly and in good shape; their respiratory and hemodynamic status were normal; they were uneventfully extubated in the operating room, monitored in the operating room for a period of time, and safely transferred to an intensive care unit once stable and fully awake.

Postoperative Care and Outcome

Only one pair benefited from advanced postoperative analgesia, including continuous peridural infusion of ropivacaine and IV morphine titration. Two pairs received acetaminophen and niflumic acid, the other babies benefited from acetaminophen sole. Complications occurred in the postoperative period in 2 babies: parietal infection in one baby, well treated by antibiotics and wound care, one baby presented an abdominal compartment syndrome which needed a 2nd surgery. Fourteen babies have survived and have been developing well since surgery. **The figure 1** showed a conjoined twins who has survived after surgery.

Death: the twin separated from a dead twin died from respiratory infection on postoperative day 4. The baby with hydrocephalus and spina bifida died from postoperative neural infection. One baby died at day 4 from multiorgan failure: preoperative echocardiography had labelled VSD what proved to be a double-outlet right ventricle and the important cross-circulation revealed during pericardial dissection was unexpected.

Table 1: Perioperative characteristics of conjoined twins in each country

	Benin	**Burkina Faso**				**Guinea**		**Niger**	**Senegal**
	Couple 1	Couple 2	Couple 3	Couple 4	Couple 5	Couple 6	Couple 7	Couple 8	Couple 9
Gender	F	F	M	F	F	F	M	M	F
Type of conjoined twins	Xypho-omphalopagus	Omphalopagus	Omphalopagus	Omphalopagus	Omphalopagus	Xypho-omphalopagus	Pygopagus	Pygopagus	Thoraco-omphalopagus
Delivery	C-section	C-section	C-section	C-section	C-section	Vaginal	Vaginal	Vaginal	C-section
Weeks of pregnancy at birth	38	39	Unknown	36	39	33	Unknown	Unknown	Full term
Total birth weight of both twins	5,200 grams	4,275 grams	4,500 grams	5,200 grams	5,000 grams	3,000 grams	3,080 grams	4,100 grams	4,300 grams
Preoperative laboratory values	CT1: ACT =40 s INR = 61% of normal CT2: ACT = 43 s INR = 51% of normal	Normal	CT1: Hemoglobin = 9,2g/ dl CT2: Hemoglobin = 11,4 g/dl Alanine aminotransferase = 90 IU/L	Normal	Normal	Normal	Normal	Normal	Normal
Echocardiography	Yes, normal	No	CT1: PFO CT2: ASD	CT1: PFO CT2: PDA, PFO	Yes, normal.	No	CT2: PFO	Yes, normal	CT2: VSD
Preoperative scanner/MRI	Yes/No	No/No	Yes/No	Yes/No	Yes/No	No/No	Yes/No	Yes/No	Yes/No
Shared organs	Liver	Liver	Liver	Liver	Liver	Liver	Sacrum, coccyx	Coccyx, penis, rectum	Liver
Associated malformations	None	None	Cardiac in both	Cardiac in Twin 2	None	None	Sexual ambiguity	Twin 1: spina bifida, hydrocephaly, single kidney Twin 2: 2 right kidneys	Elevated dia-phragmatic domes Chest skeletal abnormalities
Emergency	No	Ulcerated omphalocele	No	No	No	Pneumonia and death of CT1	No	Malaria and urinal infection in CT2	No
Age at surgery	D6	D2	M7	M11	M5	D23	M2	M3	D47

Anesthesia teams	1PA/1RA/ 2NPA	1PA/3NPA	2PA/2AR/ 1NPA	2PA/2AR/ 1NPA	3PA/2NPA	1PA/2NPA	4PA/4NPA	3PA/4NPA	4PA
Anesthesia induction	Sequential	Simultaneous	Sequential	Sequential	Sequential	Not applicable	Sequential	Sequential	Simultaneous
	Inhalational (Halothane, fentanyl)	IV (Propofol, sufentanil)	IV (Propofol, fentanyl, rocuronium)	Inhalational (Sévoflurane, fentanyl)	IV (Propofol, sufentanil, rocuronium)	Inhalational (Halothane, fentanyl)	Inhalational (Halothane, fentanyl)	IV (Propofol, fentanyl)	IV (Propofol, fentanyl)
Intubation	Easy	Impossible in CT1 Easy in CT2	Easy	Easy	Easy	Easy	Easy	Difficult	Easy
Maintenance medications	Halothane, Fentanyl	Halothane, Sufentanil	Isoflurane Fentanyl	Isoflurane Fentanyl	Isoflurane Sufentanil	Halothane, Fentanyl	Halothane Fentanyl	Halothane, Fentanyl, Atracurium	Sevoflurane, Fentanyl, Vecuronium
Monitoring	POx, PCS	POx, PCS, T	POx, ECG, NIBP, $EtCO_2$, T	Pox, ECG, NIBP, T, $EtCO_2$	POx, ECG, NIBP, $EtCO_2$	POx, PCS	POx, ECG	POx, ECG, PCS	POx, ECG, NIBP, $EtCO_2$, urinary catheter, oral temp.
	Clinical observation	Clinical observation	Clinical observation	Clinical observation	Clinical observation	Clinical observation	Clinical observation	Clinical observation	Clinical observation
Ventilation	Manual	Manual	Mechanical	Mechanical	Mechanical	Manual	Manual	Manual	Mechanical
Anesthesia time	120 mins	150 & 160 min	115 & 130 min	185 min	185 & 202 min	355 min	90 min	160 & 180 min	120 min
Outcome /Discharge	CT1 survived/D21 CT2 survived/D21	CT1 survived/D8 CT2 survived/D8	CT1 survived/D30 CT2 survived/D30	CT1 survived/D35 CT2 survived/D35	CT1 survived/D26 CT2 survived/D26	CT2 died/ D4	CT1 survived/D9 CT2 survived/D9	CT1 died/D7 CT2 survived/D82	Up to 21 days 1 survivor

Abbreviations:
AB = Antibiotic
ACT = Activated Cephaline Time
ALAT = alanine aminotransferase
AR= Anesthesiology resident
ASD = Atrial Septal Defect
ASD = ATRIAL Septal Defect
D = day

Difficult = requiring multiple attempts and placement by PA
DORV = Double Outlet Right Ventricle
$EtCO_2$ = End-Tidal CO_2 capnometry
ICU = Intensive Care Unit
INR = International Normalized Prothrombin Time Ratio
IV= Intravenous
M = month
MRI = Magnetic Resonance Imaging
NPA = Non-Physician Anesthetist
NIBP = non-invasive blood pressure
PA = Physician Anesthetist
PCS = Precordial Stethoscopes
PDA = Patent Ductus Arteriosus
PFO = Patent Foramen Ovale
POx = Pulse oximeter: all concerned countries had been provided with Lifebox pulse-oxymeters during the preceding years
RPA = Resident Physician Anesthetist

Table 2: Previous conjoint twins reported in five West Africa countries

	Benin	**Burkina Faso**	**Guinea**	**Niger**	**Senegal**
Population	**11 million**	**20 million**	**10 million**	**27 million**	**16 million**
Period	**10 years**	**15 years**	**7 years**	**5 years**	**23 years**
Recorded pairs	4 pairs, not operated	2 thoraco-omphalopagus pairs, not operated	1 omphalopagus operated in France	1 pair: failed surgery	1 thoracopagus pair operated by a local team
Description/outcome	No survivor	No survivor	2 survivors	No survivor	2 survivors
Recorded pairs			1 pair waiting for transfer abroad	1 pair: 1000-Km road transfer	
Description/outcome			No survivor	No survivor	
Recorded cases			1 Inoperable pair craniopagus		
Description/outcome			2 survivors		
Total concerned babies	8	4	6	4	2
Total survivors	0	0	4 (2 inoperable)	0	2

Figure 1: A conjoins twins who has survived.
Photo A: Conjoins twins before surgery; photo B: Conjoins twins after surgery

DISCUSSION

The largest international epidemiological study of conjoined twins reported so far concerned 383 sets of CTs born among a total of 26,138,837 births, a global prevalence of 1,47 per 100,000 births [1]. India and Africa provided no data while high frequencies of 1/14,000 have previously been reported in Uganda [2]. Reports of live CTs are rare in Africa. The present series matches the two-to-one global predominance of female CTs. Cesarean delivery is the accepted standard for anticipated CTs delivery [3, 4], but three vaginal deliveries occurred in this series.

The reported cases in this study were not the first symmetrical CTs brought to the knowledge of the authors and Table 2 presents those previous cases. At

least 3 pairs died awaiting international transfer. These cases represented a recollection and not national databases, and some cases may never have been transferred in a hospital. This series confirms that regular follow-up of pregnancies and medically assisted birth are still rare occurrences in rural environments, perinatal and hospital hygiene is rudimentary, nosocomial infections are frequent.

Surgical separation is best performed between the 4th and 11th months of life [5]. In this series three separations were before the 4th month because of emergencies, three separations were run at the right moment and three early because of the social pressure, CTs were not accepted. Anesthesia for CTs separation remains a challenge, even in technically advanced countries, and in Sub-Saharan Africa, CTs were most often referred abroad for surgery. But budgetary considerations and the need to find a foreign accepting hospital can lead to long and sometimes fatal delays. Perioperative logistics and organization are complex: two surgical and anesthesia teams must work simultaneously in a single operating room; all material must be available in duplicate [3-8]. In these countries emergency surgeries constitute the majority of surgical activity. Due to the shortages of materials and personnel, scheduling simultaneous complex interventions is a difficult endeavor for even larger hospitals, and urgent or emergent interventions for CTs is problematic. The complexity of possible defects requires thorough preoperative anatomical workup with occasionally the need to quantify and localize cross-circulation [3-6]. Preoperative imaging failed to provide the needed information to most operative teams, because technical means to assess the anatomy of CTs may be unavailable, not located in public hospitals, or unaffordable. Relative blood shortage is the rule in these countries but for these surgeries blood was made available in accordance with the blood banks. Last but not least, the uncertain and irregular supply of some necessary devices such as blood pressure cuffs for neonate and infants, for anesthesia and postoperative pain management is a significant issue; and families often cannot afford to pay for even essential drugs such as antibiotics. In this series, only one pair received adequate analgesia. The mortality in this series, 4 in 18 babies, is a notable progress compared with our historical data. However, both series concerned rather uncomplicated CTs: it is possible that more complicated cases never reached a hospital. Immediate postoperative care was provided by anesthesiologists in the operating room, for lack of recovery rooms. One hospital lacks a specific pediatric intensive care unit and existing ones feature great shortages (material, drugs). Finally, fundamental reforms are needed to provide financial support for poorest families confronted to the need for surgery.

However, more than technical means, knowledge and skills were crucial to meet the challenge. All coauthors graduated from local training programs in Benin, Burkina Faso and Senegal, including one year residency in Europe or North Africa, and got complementary training in pediatrics anesthesia or have some regular practice of it. Unsophisticated manual ventilation techniques, rigorous clinical monitoring and drugs were resorted to. The essential safety provided by the LIFEBOX* pulse-oximeters is worth mentioning.

CONCLUSION

CT separation surgery still constitutes an extraordinary challenge in LMICs, but well-trained anesthesiologists, a multidisciplinary approach and international contacts can lead to safe performance of complex neonatal surgeries. However, countries need to improve methods for sourcing inexpensive and basic monitoring equipment and pharmaceuticals to reach acceptable safety conditions.

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The authors confirm that all authors have made substantial contributions to all of the following:

- The conception and design of the study, or acquisition of data, or analysis and interpretation of data.
- Drafting the article or revising it critically for important intellectual content.
- Final approval of the version to be submitted.
- Sound scientific research practice

The Authors Further Confirm That:

- The manuscript, including related data, figures and tables has not been previously published and is not under consideration elsewhere
- No data have been fabricated or manipulated (including images) to support our conclusions
- This submission does not represent a part of single study that has been split up into several parts to increase the quantity of submissions and submitted to various journals or to one journal over time (e.g. “salami-publishing”).

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The Authors Declare no Conflict of Interest

"The author/s declare that this submission is in accordance with the principles laid down by the Responsible Research Publication Position Statements as developed at the 2nd World Conference on Research Integrity in Singapore, 2010."

"All procedures were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

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