



Case report

Use of local perforator flaps for closure of a thoraco-omphalopagus conjoined twin defect after separation during the COVID-19 pandemic

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ABSTRACT

Introduction: Conjoined twins are a rare medical phenomenon that poses unique challenges for surgeons. Separation of conjoined twins involves multidisciplinary teamwork, complex medical management and surgical planning, and multi-stage operations and often still has a high mortality and morbidity rate. In the times of the COVID-19 pandemic, separation of conjoined twins pose even greater challenges. Aiming for the best outcome possible, while minimizing the risk of COVID transmission and ensuring the safety of the personnel, is paramount. This case report presents thoraco-omphalopagus twins who were successfully separated at 4 months of age. The preoperative planning, operative details, postoperative follow-ups, and outcomes are discussed.

Methods: The absence of a tissue expander and the inability to acquire it due to travel restrictions from COVID-19 further complicated the management on this patient. A Routine Polymerase Chain Reaction (PCR) swab test was performed on the patients and personnel. Standardized personnel protective equipment (PPE) was worn during ward and surgical care. After separation of the twins by cardiothoracic and pediatric surgeons, one twin underwent immediate skin closure using a double keystone perforator island flap and a lower abdominal perforator flap. Due to extensive defects, closure was delayed for the second twin. After a series of dressing changes, eventually local perforator flaps could be raised to close the defect using staged tension sutures and skin grafts for secondary defects.

Results: Both twins were discharged with no significant morbidity, and no personnel were exposed to COVID-19 infection during the management.

Conclusion: Preoperative coordination and planning, multidisciplinary effort, adherence to screening protocols for COVID, and strict use of standardized PPE all contributed to the successful separation of thoraco-omphalopagus conjoined twins during the COVID-19 pandemic.

1. Introduction

Conjoined twins are a rare medical phenomenon that occurs as a result of monoamniotic and monochorionic gestation, when two identical individuals are joined by part of their anatomy and share one or more organs [1]. The incidence of conjoined twins ranges from 1:50,000 to 1:100,000 live births [2, 3]. Based on the classification proposed by Spencer et al [4], thoraco-omphalopagus conjoined twins join from the thoracic region to the umbilical with varying degree of fusion of heart, liver, and gastrointestinal tract. Separation surgery of conjoined twins still results in high mortality and morbidity [5].

In the times of COVID-19 pandemic, especially in a developing country where the cases are still increasing and peak of the curve is not yet reached [6], the risks of disease transmission both to the patient and to the surgical teams are multiplied [7, 8]. Furthermore, a shortage of personal protective equipment, the inability to acquire tissue expanders that are widely used for conjoined twin separation, and the limited capacity of intensive care units and hospital resources also pose even greater challenges in the management of conjoined twins. Most reports on conjoined twins have mainly focused on the multidisciplinary teamwork and outcome of the separation [1, 3, 5, 9, 10, 11, 12], but only a few reports have described the design and use of flaps for defect closure after separation [13, 14].

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In this report, we present a case of a set of conjoined twins undergoing separation surgery. Our focus in this study is the use of local perforator flaps of the thoracoabdominal as soft tissue coverage of the defect after separation. We also present mitigation strategies for performing conjoined twin surgeries to prevent the risk of COVID transmission among the surgical teams, while still aiming for the best and safest outcome possible for the patients. The separation was a success technically, although one of the twins only survived a few weeks after being discharged.

2. Case presentation

2.1. Patient information

A set of female twins, weighing 3.5 kg at birth, were born prematurely. The twins were joined from the chest to abdomen. The caesarean delivery was performed in the regional hospital, and the twins were directly referred to our tertiary hospital and admitted to the neonatal intensive care unit (NICU). A multidisciplinary team consisting of pediatricians, anesthesiologists, pediatric surgery team, cardio-thoracic surgery team, and plastic surgery team was assembled to plan the separation. The cost for the separation surgery and inpatient care were all covered by the hospital, considering this was a really rare case. Informed consent was obtained from the patients for publication of the case report details. The study was performed in accordance with the principles of the Declaration of Helsinki.

2.2. Clinical findings

The twins were joined from the lower anterior thorax to the level of the umbilicus. The fusion was at the midline, and the area of skin on the lateral parts of the twins were relatively symmetrical. The CT scan imaging revealed that they shared a rib cage with 2 pairs of lungs and 2 separate hearts, and a shared liver, but with separate GI tracts (Figure 1). Other than the fused parts, both babies showed normal anatomical presentations and normal extremities. One of the twins had smaller body proportions and was named Baby 1.

After admission to the NICU, Baby 1 developed respiratory distress and was put on a ventilator. Both babies were screened for COVID-19 and Baby 1 was found positive for COVID-19. After a series of hemodynamic surges and ventilator-associated pneumonia, Baby 1 was finally stable at the age of 5 months, and the Polymerase Chain Reaction (PCR) test for COVID-19 was negative. To avoid further instability and deterioration in

the hemodynamics of Baby 1, the separation surgery was scheduled at this period. See Table 1 for the timeline of multiple surgery on both babies. Both babies showed positive results for a rapid COVID-19 test during preoperative screening, ten days after the separation surgery. This made further management of the patients more complicated because they had to be moved to the COVID ward before finally testing negative twice by PCR for COVID-19. They were then transferred back to the PICU in the non-COVID ward.

2.3. Therapeutic intervention

Initial preoperative screening tests for COVID-19 using the PCR test twice for each baby from oropharyngeal swabs and endotracheal swabs resulted in all negative tests for the twins preoperatively, so the first surgery to separate the babies was performed. The incision of the skin was done in a straight-line manner. The separation surgery was performed by a pediatric surgery team and a cardio-thoracic surgery team to separate the liver and rib cage. The liver was separated using cavitron ultrasonic surgical aspirator, a branch of portal vein which cross over the separation line was divided and ligated, the parenchyma was then sutured with polypropylene. The surgery was then continued by the plastic surgery team.

The defect on Baby 2 was 9×15 cm, with the chest and abdomen covered by mesh. Perforators from the lateral abdomen were identified using hand-held Doppler ultrasound. Bilateral keystone design perforator island flaps (KDPIF) were designed, with the area of the perforators included in the design. The flaps were incised and elevated subfascially. More than fifty percent of the keystone design perforator island flaps were undermined for mobility, and the flaps were mobilized to close the thoracic and upper abdominal defect with an omega design, with a third flap from the lower abdominal flap advanced cranially to close the remaining abdominal defect. All the defects from Baby 2 were able to be covered in the separation surgery (Figure 2).

After the separation, the abdomen of Baby 1 was covered with mesh. A part of the chest wall was reconstructed with miniplates and the heart was covered with Gore-tex®. The defect on Baby 1 was 15×25 cm. Because of the unstable hemodynamics after separation and the high possibility of developing abdominal compartment syndrome if the skin closure was performed, the team decided to delay the skin closure of Baby 1.

Three days after the separation, Baby 1 was scheduled for surgery to close the defect. Perforators from lateral thoracic area were identified with handheld Doppler ultrasound, and then the KDPIF was elevated in

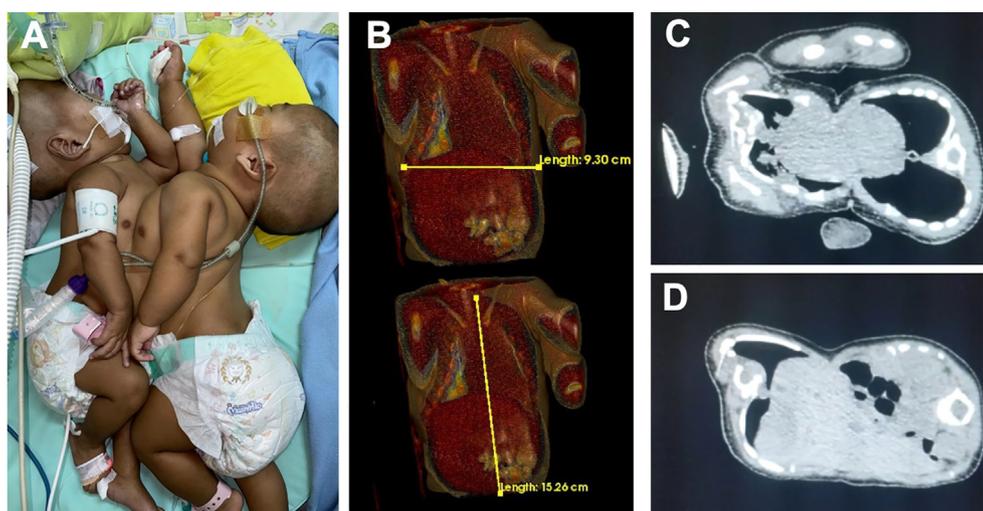


Figure 1. A, The twins before the separation surgery, Baby 1 is on the left, Baby 2 on the right. B, CT-generated model to determine the size of the defects pre-operatively. C, CT image from the thoracic level showing the shared rib cage with 2 pairs of lungs and 2 separate hearts. D, CT image from abdomen level, showing the shared liver with separate GI tracts.

Table 1. Timeline of surgery of the defect closure after separation.

Timeline	Baby 1	Baby 2
July 13, 2020	Separation surgery by the pediatric and cardio-thoracic surgery team Baby 1: Temporary closure of defect using Gore-tex® and mesh. Soft tissue coverage was not yet performed Baby 2: Double keystone perforator island flap and lower abdominal flap for soft tissue coverage	
July 16, 2020	Closure of the thoracic defect and partial abdominal defect using double keystone perforator island flaps, an abdominal flap, and NPWT	
July 24, 2020	Debridement and NPWT changes	
July 25, 2020		Debridement and resuturing of the flap, and applying NPWT*
July 28, 2020		Debridement and resuturing of the flap, and applying NPWT*
August 4, 2020	Re-elevation of the flap, dynamic suturing, and NPWT	
August 12, 2020		Debridement and primary closure
September 7, 2020	STSG for the secondary defects	

NPWT: Negative-pressure wound therapy, STSG: Split-thickness skin graft.

* Period of suspected COVID-19 re-infection.

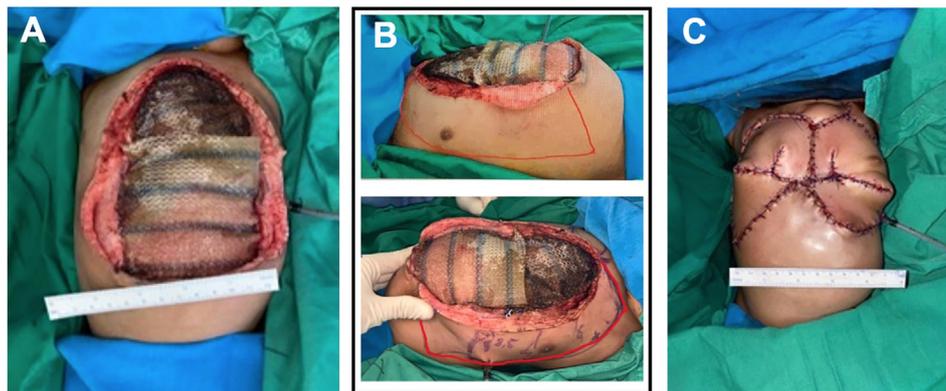


Figure 2. Defect closure of Baby 2. A, The defect with the size of 9 × 15 cm. B, Design of the KDPIF with the dot marking indicating the location of the perforator based on preoperative Doppler ultrasound examination. C. Postoperative appearance.

an omega manner to close the defect mainly to cover the chest with the exposed Gore-tex® and miniplates. A third perforator flap from the lower abdomen was raised to cover the lower abdominal area. After inseting the flap, a defect remained on the abdomen with a size of 6 × 10 cm

(Figure 3). The remaining defect was covered with negative pressure wound therapy (NPWT).

A week later, baby 1 subsequently was noted to have purulent discharge from the thoracic and upper abdominal region, so we decided

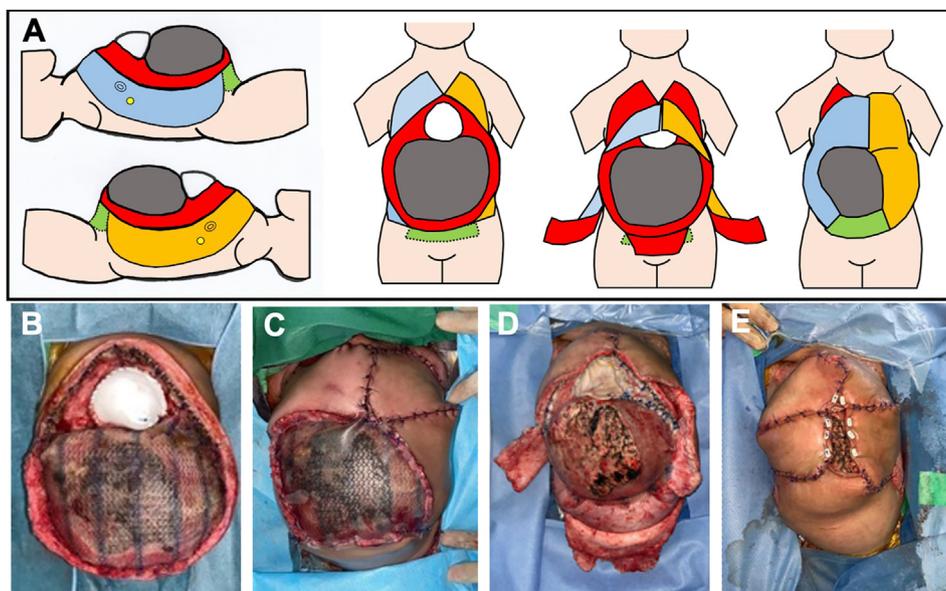


Figure 3. Defect closure of Baby 1. A, The schematic illustrations of bilateral KDPIF and abdominal flap (Blue: right KDPIF, orange: left KDPIF, green: abdominal flap, yellow dot: perforator). B, The defect with the size of 15 × 25 cm. C, Postoperative appearance. The remaining defect was covered with NPWT. D, Re-elevation and resuturing of the flaps. E, Application of dynamic sutures to increase skin expansion.

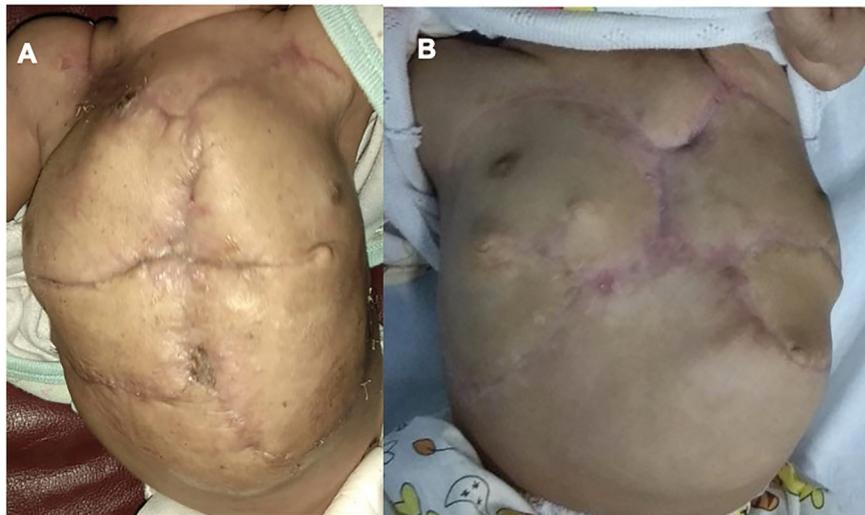


Figure 4. A, Baby 1 during the follow-up period; the wounds were epithelialized. Baby 1 passed away two weeks after being discharged. B, Baby 2 during the follow-up period; the wounds were epithelialized.

to open several sutures covering the thoracic region and perform debridement. We then covered the defect with 100 mmHg continuous-mode NPWT. After 4 days, we evaluated the wound and found that some slough still remained in the thoracic region, so we decided to continue the NPWT.

Ten days after the separation, a rapid antibody COVID test was performed on both babies and both showed positive results. The babies were then immediately transferred to the COVID ward according to hospital policy. Fortunately, based on the contact tracing, no other patients or staff were positive for COVID-19.

Baby 1 had one NPWT change procedure in the COVID operating room (OR). Baby 2 developed tip necrosis and dehiscence on the thoracic region at the right upper limb of the keystone perforator island flap and went to the operating room for debridement. During surgery, we found that the mesh was covered with slough, so several sutures were opened, NPWT was applied to the wound, and the mesh was taken out in the infected area. Three days later, Baby 2 went to the OR again for NPWT change and debridement. The thoracic region was still sloughy, so we left the wound open and applied NPWT. Both babies stayed in the COVID ward for 1 week until two swab PCR tests showed negative results. They were then transferred to the pediatric intensive care unit (PICU). Both babies showed improvements in their ventilation problems.

After a series of NPWT changes, Baby 2 underwent the last surgery 1 month after the separation. The remaining defects on the midline were closed primarily. The defect was epithelialized and Baby 2 was discharged 10 days later.

For Baby 1, dynamic sutures were applied to give the expansion effect of the skin flap (Figure 3). After 2 procedures of tightening the dynamic sutures, and improvement in the general condition as well as the ventilation of Baby 1, we performed surgeries to cover all the remaining defects. We re-elevated the lower limb of the right keystone perforator island flap and the lower abdominal perforator flap to cover the remaining defects (Figure 3). Finally, almost 2 months after the separation, Baby 1 underwent the last surgery with a split thickness skin graft (STSG) to cover the secondary defects on the flank. All the defects were epithelialized and the patient was also discharged 1 week later (Figure 4).

All the surgeries were performed with as minimal personnel as possible to reduce the risk of COVID transmission. Every surgical workforce must wear standardized personnel protective equipment according to the level determined by the hospital. The use of N95 masks and eye protectors was mandatory for surgery in the non COVID OR, while additional hazmat and boots were used in the COVID OR.

2.4. Follow-up and outcomes

During the follow up period, the twins showed good healing of the skin on the chest and abdomen and significant gains in the body weight. Unfortunately, two weeks after being discharged, Baby 1 developed symptoms of pneumonia, with fever and shortness of breath. Before having the chance to be brought back to our unit, Baby 1 passed away. The death was suspected to be related to COVID-19 but no autopsy was performed due to the cultural beliefs of the parents. Currently, Baby 2 is healthy, has normal growth, and is still under routine follow-up. The risk for ventral hernia will be assessed during follow-up and abdominal wall reconstruction will be done accordingly one until three years after the separation surgery [15].

3. Discussion

Conjoined twins are regarded as monozygotic, monochorionic twins of the same sex with an identical chromosomal pattern [2, 4]. One of the most common types of conjoined twins is the thoraco-omphalopagus type, which accounts for 75% of these cases [16]. Surgical separation of these twins has been always a challenge and may require novel techniques. One of the challenges of the separation is tissue coverage of the defect after separation. Most cases of thoraco-omphalopagus conjoined twins result in large defects of the anterior chest and abdomen [5, 9].

Numerous strategies for soft tissue coverage have been reported. A variety of skin grafts and skin flaps and the use of tissue expander procedures have been described. Skin grafts, however, require an intact body wall for use in closure [17]. Skin flaps have been successful and reliable for tissue coverage [13, 14], as well as pedicled flaps such as latissimus dorsi flap and superficial circumflex iliac artery perforator flap [18, 19]. Tissue expanders have arisen as an optimal means of achieving wound closure in conjoined twin separation, with the first reported success in 1986 [20]. However, the use of tissue expanders has its own disadvantages, namely the need for multiple procedures and the possibility of the expander creating pressure into the abdominal cavity and causing intraabdominal compartment syndrome [5].

In this study, we used local perforator flaps from the lateral chest and abdomen area to close the defects on the anterior chest and abdomen. In this case, we used perforators from the lateral thoracic arteries. We used the hand-held doppler ultrasound to estimate the location of the perforator. A color doppler ultrasound would be more precise but it is not a standard operational procedure in our hospital and we tried to minimize the number of person involved in the management of the patients. We

postulate that perforators from this region are a reliable source of vessels for the flaps. Based on the studies by Hocaoglu and Aydin [21], Offman et al. [22], and Taylor et al. [23], the perforators in the flank region arise from the intercostal, lumbar, lateral thoracic, and thoracodorsal arteries. We opted for a keystone island flap design to allow mobility and reduce tension. The keystone flap was first introduced by Behan [24] as a multi perforator advancement flap with the advantages of providing a reliable, safe flap harvest that can minimize donor site morbidity and obviate the need for microsurgical techniques. To the best of our knowledge, this is the first report of the use of a keystone perforator island flap for defect closure after conjoined twin separation.

Because of the extent of the defects and the relatively small size of the baby compared to the defect, the flaps could not cover all the defects in Baby 1 in a single stage operation. We used dynamic sutures for defect closure, as stated by Taylor et al. [25], where the use of dynamic sutures as a delayed primary closure would give an expansion effect because of the extensibility and elasticity of the skin.

Despite the successful separation and good closure and healing of the skin, Baby 1 did not survive beyond two weeks after the discharge. Pneumonia due to COVID-19 infection was suspected as the cause of the worsening condition of Baby 1. Furthermore, prior to separation, Baby 1 had experienced pneumonia and was on a ventilator for a period of time; therefore, the respiratory condition of Baby 1 had not been as good as that of Baby 2. Neither PCR swab test or antigen swab examination was done to baby 1 due to socioeconomic issues, but during the postoperative visits, baby 2 and the mother also had a positive antigen swab test of COVID-19 with no respiratory symptoms.

4. Conclusion

This report demonstrated that a local perforator flap could be considered as a preferred method for reconstructing the defect after conjoined twin separation, even in settings where tissue expanders are not available. With careful and detailed preoperative planning, precise marking of the flap, meticulous dissection, and use of dynamic sutures, closure of the defect can be achieved.

Lastly, every surgery performed in the times of the COVID pandemic must aim to reduce the risk of transmission and ensure the safety of the patients and of the surgical team. These strategies include strict screening protocols, minimizing the number of surgeons and surgical staff involved in the surgery, adherence to the use of standardized PPE, and shortening the duration of surgery, while still aiming for the best outcome possible.

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The authors declare no conflict of interest.

Additional information

No additional information is available for this paper.

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