Conjoined Twins : Surgical Separation in 11 Cases

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Abstract

Eleven pairs of symmetrically conjoined twins underwent surgical separation at the Queen Sirikit National Institute of Child Health. Six were omphalopagus, 4 were thoracopagus and 1 was pygopagus. Eight were female and 3 were male. Three pairs were separated on emergency or semi-emergency bases, and the remaining 8 pairs were separated electively at an older age.

Of the 3 pairs who had early emergency separations, one pair, whose combined birth weight was only 2,500 g, underwent emergency separation at the age of 44 days after the death of one twin. The second twin also expired one hour after the separation. In the remaining 2 pairs, early separation was done because of the deterioration of one twin due to complex cardiac anomalies. In both cases, the infants with cardiac anomalies expired but the others survived the separation satisfactorily.

In one pair of thoracopagus conjoined twins, one twin had cyanotic cardiac anomalies. They were electively separated at the age of 2 years and 9 months. The twin with cardiac anomalies expired 2 hours after surgery, but the other survived the separation satisfactorily.

In the remaining 7 pairs who underwent elective separations, both twins of each pair survived the separation satisfactorily. However, one twin expired unexpectedly 10 days after the separation.

Key word : Conjoined Twins, Siamese Twins, Thoracopagus, Omphalopagus, Pygopagus, Separation

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Conjoined twinning is one of the rarest congenital anomalies. Whenever it appears in newspapers, it is always received with emotion and fascination by the public as well as physicians. Because of its rarity, the terminology regarding the types of malformation is difficult for most physicians to remember. The most famous conjoined twins were Eng and Chang Bunker, who were born in Thailand in 1811. They were called "Siamese twins". Since then, the name "Siamese twins" has been the best known term for all kinds of conjoined twins. Surgical separation of conjoined twins is one of the greatest challenges to surgeons.

Several classifications of conjoined twins have been suggested[1-3]. Classification by Potter[1], which has been the most widely used classification, divides conjoined twins into 2 main groups: diplopagus (both twins are equal and symmetrical to each other) and heteropagus (unequal and unsymmetrical conjoined twins) (Table 1). Diplopagus with complete or near complete twins are more common than other groups. Most of the recent textbooks of pediatric surgery limit the discussion of this topic in detail to this group only and separate omphalopagus from thoracopagus[4-7].

Thoracopagus conjoined twins, by definition, are joined at the anterior chest and upper abdominal walls down to the level of the umbilicus (Fig. 1). *Omphalopagus* or *xiphopagus* twins are joined at the upper anterior abdominal wall. *Ischiopagus* twins are joined at the lower anterior abdominal wall and perineum. *Pygopagus* twins are joined at the buttock and perineum in a back-to-back position. *Craniopagus* twins are joined at the head. Only these 5 types of conjoined twins are included in the present report.

Surgical Separation at Queen Sirikit National Institute of Child Health

A total of 11 cases of conjoined twins were operated upon at the Queen Sirikit National Institute of Child Health (QSNICH) (Table 2). Nine cases have previously been reported in detail[8-11]. Additional 2 cases will be presented in detail in this report.

Case 10

These female conjoined twins were born by cesarean section at Samronge Hospital, Samut Prakan Province on November 16, 1995, with a combined birth weight of 4,050 grams. They appeared to be omphalopagus conjoined twins with fusion at the epigastrium (Fig. 2). Auscultation of the chest revealed normal heart and respiratory sounds in both twins.

Chest film was normal. Ultrasound of the abdomen revealed fusion of the livers only. The kidneys, bladder, spleen and gall bladder were all normal in both infants. Intravenous pyelography (IVP) of each infant showed no cross visualization of the kidneys and bladder. Voiding cystourethrography (VCUG) showed no connection of the lower urinary tract of the twins. A long gastro-intestinal contrast study showed no connection between the gastro-intestinal tract of the twins.

Other than occasional respiratory tract infections, the twins appeared healthy and did not have other health problems.

Table 1. Classification of conjoined twins (modified from Potter[1]).

I. Diplopagus (Both twins are equal and symmetrical)
- (1) Each twin is complete or nearly complete
  - 1. Thoracopagus, omphalopagus (xiphopagus)
  - 2. Ischiopagus
  - 3. Pygopagus
  - 4. Craniopagus
- (2) Each twin is not nearly complete
  - 1. Duplication originating in the cranial region.
    - 1.1 Monocephalus (single head with partial duplication of facial structures)
    - 1.2 Dicephalus (two heads with lateral fusion of the trunks)
  - 2. Duplication originating in the caudal region (Dipygus)
    - 2.1 Monocephalus - tripus dibrachius
      - tetrapus dibrachius
    - 2.2 Cephalothoracopagus
  - 3. Duplication of both cranial and caudal regions (Dicephalus dipygus)

II. Heteropagus (Unequal and asymmetrical conjoined twins)

Table 2. Surgical separation of conjoined twins at QSNICH.

Case no.	Sex	Combined birth weight (g)	Type of twins	Age at separation	Date of separation	Indication for separation	Shared organs	Result	
								Twin 1	Twin 2
1.	F	?	OMPH	14 mo.	Jan 20, 1956	Elective	- Xiphoid - Liver bridge	Survived	Dead, 10 days post op
2.	F	3,700	OMPH	19 mo.	Dec 22,1962	Elective	- Xiphoid - Liver bridge	Survived	Survived
3.	F	3,950	OMPH	17 mo.	1964	Elective	- Xiphoid - Liver bridge	Survived	Survived
4.	F	2,500	OMPH	44 days	Aug 13, 1963	Emergency (Death of one twin)	- Xiphoid - Liver bridge	Dead, 1 hour after separation	Dead before separation
5.	F	4,720	THOR	2 yr. 9 mo.	Oct 18, 1983	Elective	- Sternum - Pericardial cavity - Bridging channel connecting atria - Diaphragm - Liver bridge	Survived	Dead, 2 h post op (Complex Cardiac anomalies)
6.	F	4,470	THOR	4 mo.	Apr 24,1989	Elective	- Sternum - Pericardium - Diaphragm - Liver bridge	Survived	Survived
7.	M	4,040	OMPH	6 days	Apr 27, 1900	Semi-emergency	- Xiphoid - Liver bridge	Survived	Dead, 18 days post op
8.	M	4,800	THOR	29 hours	Apr 8, 1993	Emergency (Deterioration of one twin)	- Sternum - Pericardium - Diaphragm - Liver bridge	Dead in OR	Survived
9.	M	4,680	THOR	5 mo.	Aug 15, 1995	Elective	- Sternum - Pericardium - Diaphragm - Liver bridge	Survived	Survived
10.	F	4,050	OMPH	3 mo.	Feb 6, 1996	Elective	- Liver bridge	Survived	Survived
11.	F	4,250	PYGO	2 mo.	Feb 17, 1999	Elective	- Vagina - Rectum - Coccyx	Survived	Survived

THOR = Thoracopagus, OMPH = Omphalopagus, PYGO = Pygopagus

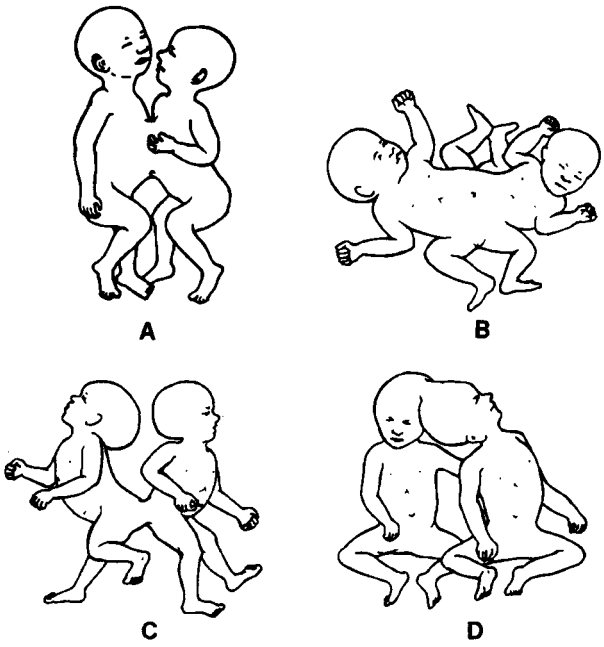

Fig. 1. A) Thoracopagus, omphalopagus, xiphopagus. B) Ischiopagus. C) Pygopagus. D) Craniopagus.

Surgical separation was undertaken on February 6, 1996. The peritoneal cavity of each twin was connected to each other through a 2 cm opening, and there was a connecting liver bridge between the anterior surface of the lateral segment of the left lobe of the liver of each baby (Fig. 3). After an application of 2 pairs of vascular clamps on the liver bridge, the liver bridge was divided between the clamps with electric cautery. Each stump of the liver bridge was sutured with multiple 2-0 chromic catgut sutures. Closure of the abdominal wall was accomplished with ease on each twin. The wounds healed satisfactorily (Fig. 4). They grew normally and appeared normal when last seen at the hospital at the age of 10 years.

Case 11

These female conjoined twins were born by cesarean section at Rajavithi Hospital on December 2, 1998. Twin pregnancy was diagnosed prenatally by fetal heart sound auscultation and prenatal ultrasound, but the conjoining was not recognized by the investigation. The gestational age at birth was believed to be 36 weeks by date. The combined birth weight was 4250 grams.

Physical examination on admission to QSNICH revealed a female pair of pygopagus conjoined twins (Fig. 5, 6). The vagina of each twin was joined together at the vestibule. There was only one anal opening that was located at one side of the conjoined vaginae (Fig. 7). The corresponding location in the opposite side did not have an opening.

Abdominal film showed an unremarkable gas pattern in both twins. Lateral view of the pelvis showed no bony fusion between the sacrum of each twin (Fig. 8). Ultrasound of the abdomen showed normal liver, gall bladder, bile ducts, pancreas, spleen, kidneys and urinary bladder in each twin. Barium enema (BE) showed a short common channel of lower rectum with separate upper rectums and sigmoid colons (Fig. 9). The long gastro-intestinal contrast study showed normal upper gastro-intestinal tract and small intestine. IVP and VCUG showed a separate, normal urinary tract in each twin.

Both appeared healthy otherwise. Surgical separation was done on an elective basis on February 17, 1999. The common channel of the rectum was noted to be only about 1.5 cm long. Separation of the soft tissue and cartilaginous coccygeal fusion was done and the reconstruction of the anorectal canal was undertaken for each twin without undue difficulty. Post operative course was uneventful.

Both twins required a revisional anoplasty about one month after the separation because the anal opening appeared to be too close to the vagina. Both

Fig. 2. Case 10. Omphalopagus conjoined twins. Fusion was limited to the epigastrium only.

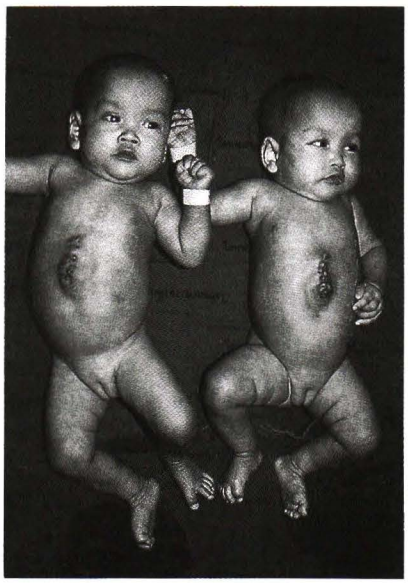

Fig. 4. Case 10. Complete healing of the incision of both twins (seven days after surgery).

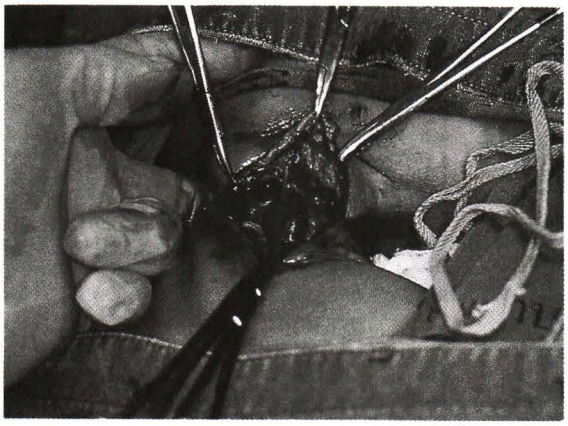

Fig. 3. Case 10. Communication between the peritoneal cavity of each twin, and the liver bridge connecting the anterior surface of the lateral segment of the left lobe of the liver of each twin.

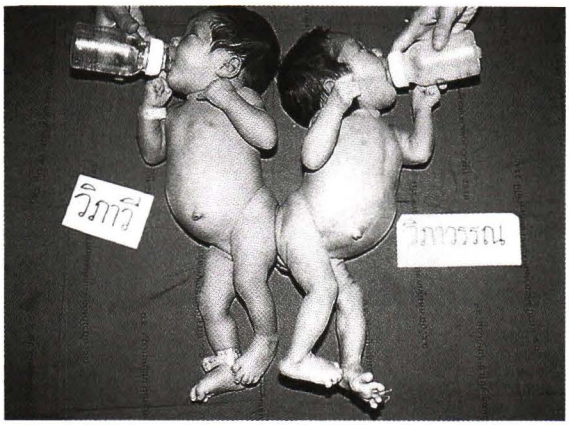

Fig. 5. Case 11. Pygopagus conjoined twins.

had occasional constipation that required occasional laxatives. They grew normally and looked healthy on their recent visits to the hospital about 3 years after surgical separation.

DISCUSSION

Conjoined twins are believed to be the result of incomplete cleavage of the embryo at approximately 2 weeks of gestation[1,5-7]. The true inci-

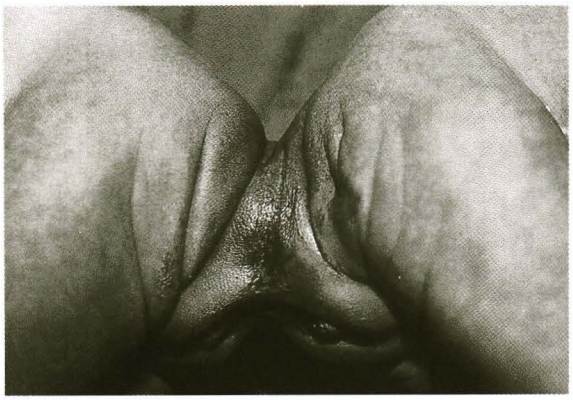

Fig. 6. Case 11. Close - up lateral view of the fusion of the buttock and perineum.

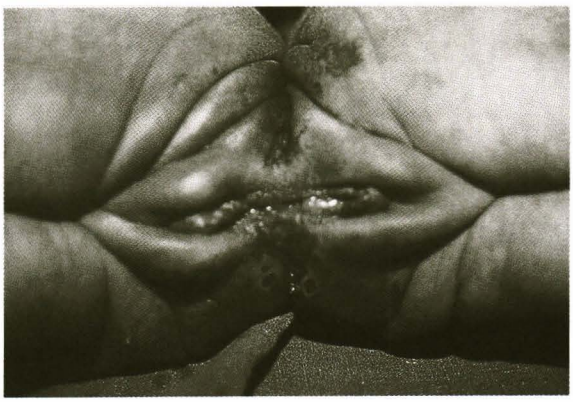

Fig. 7. Case 11. Fusion of the external genitalia at the vestibule of the vagina. Only one anal opening (arrow head).

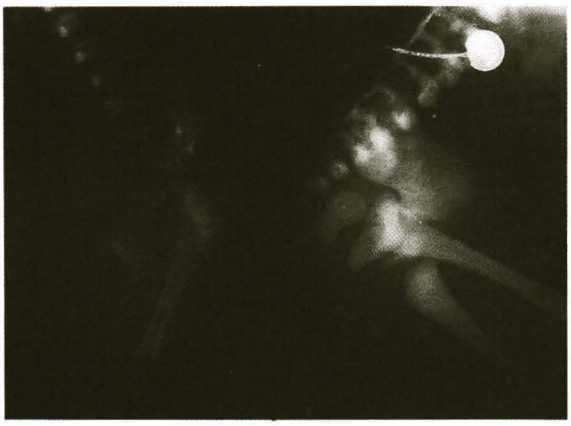

Fig. 8. Case 11. Lateral view of the pelves showed no bony fusion of the sacrums.

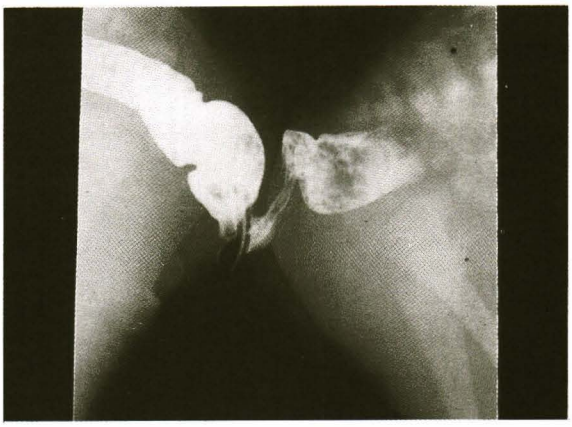

Fig. 9. Case 11. BE showed a short common channel of the lower rectum.

dence of conjoined twins is hard to ascertain. However, it has been estimated to be from one in 50,000 to one in 200,000 births[1,4,7,12-14]. The incidence in South Africa may be slightly higher[12,15,16]. A good number of the conjoined twins are stillborn or expire shortly after birth. Most reported cases in the literature appeared to be symmetrically conjoined twins of the complete or near-complete variety. Occurrence of these anomalies in females is about 2 to 3 times that in males[4-7]. According to a collective review of 117 cases by Tartuffi[17], about 73 per cent were either thoracopagus or omphalopagus, 19 per cent were pygopagus, 6 per cent were ischiopagus and 2 per cent were craniopagus conjoined twins. Thoracopagus are found more frequently than omphalopagus conjoined twins[4-7].

Of the 11 cases of surgical separation in the authors' series, 6 were omphalopagus, 4 were thoracopagus and 1 was pygopagus. This should not be interpreted that omphalopagus conjoined twins were found more commonly than thoracopogus in our hospital. It simply reflects the fact that omphalopagus twins

are more feasible for separation than thoracopagus twins. Several thoracopagus twins were taken care of in our hospital, but cardiac fusion made the separation unfeasible. They, therefore, are not included in the present report. It has been estimated that conjoining of hearts in some extents may be present in about 75 per cent of all the thoracopagus twins[4-6]. To the best of our knowledge, a successful separation with long-term survivors has never been reported in twins with ventricular fusion. Few cases may temporarily survive the separation procedure, with a sacrifice of one twin, but inevitably succumbed eventually because of complex cardiac anomalies[7]. In case no. 5, who had atrial connection, one twin survived the separation but the other expired because of cardiac anomalies. Similar success has been reported by Synhorst et al[18].

Besides the authors' cases, 6 additional cases of separation of conjoined twins at other hospitals in Thailand have been reported so far. Three were omphalopagus[19-21], two were thoracopagus[21,22], one was ischiopagus[21,23], and one was pygopagus [24]. Most reports in the literature were single case reports. Only a few communications have reported more than a few cases from the same institutes. These included the experience from Philadelphia of 18 cases, of which 13 were separated[7], and the experience of a Cape Town group of 14 cases, of which only 10 underwent surgical separation[16]. Hoyle[25], in a collective review of all 167 cases of surgical separation that had been reported in the world literature up to December, 1987, found that 29 per cent of those separated cases were thoracopagus, 25 per cent omphalopagus, 20 per cent ischiopagus, 16 per cent craniopagus and 10 per cent were pygopagus.

In recent years, prenatal ultrasonographic study has become a common practice in obstetrics in order to predict the fetal gender before delivery. Conjoined twins should be diagnosed prenatally by this technique. However, due to the rarity of this entity and the lack of radiologists' experience, conjoined twins may not be recognized during the study. At least 5 of the presented cases had prenatal ultrasonography. Twinning was diagnosed in all 5 cases, but the conjoining nature of the twins was not diagnosed in any of the cases. Criteria of ultrasonographic diagnosis in conjoined twins have been well described[26,27]. If this condition is diagnosed prenatally in small hospitals, the mother should be referred to a larger hospital, where facilities for appropriate obstetric management and neonatal intensive care are available. Of the 11 cases in the present series, 6 pairs were born by cesarean section and the remaining 5 pairs were born by vaginal delivery. Ultrasonographic diagnosis of conjoined twins have been reported to be possible as early as 12 weeks[28]. Prenatal echocardiography has been shown to correctly diagnose major cardiac anomalies in thoracopagus conjoined twins[29]. Obstetric dilemma in the management of the pregnancy is beyond the scope of discussion here. However, cesarean section appears to be the safest method of delivery for conjoined twins.

At our hospital, neonates with conjoined twinning are usually managed initially in the neonatal intensive care unit (NICU). Some of them may require endotracheal intubation and ventilatory support. Cardiopulmonary assessment is usually done early in order to prognosticate and determine the feasibility of surgical separation. Unless there is an indication for an urgent surgical separation, the procedure is preferably postponed until the optimal age is reached. The optimal age for separation is 3-12 months[4,5,7,13,14,25,30,31]. While awaiting elective surgical separation, investigations have to be done to determine the anatomical situation of both twins before a decision regarding separatability can be made. In recent years, ultrasonography and computed tomographic scan (CT scan) have proved to be very useful and have become essential for almost every case. Angiographic studies and magnetic resonance imaging (MRI) should be done in only selected cases of ischiopagus and craniopagus. Magnetic resonance angiography (MRA) may replace MRI in some cases. Echocardiography should be done in every case in which cardiac anomaly is suspected. Genitourinary tract should be evaluated with IVP and VCUG. Gastro-intestinal tract should be evaluated with BE and a long gastro-intestinal contrast study. In some cases, DISIDA scan may be done to evaluate the hepatobiliary tree.

Three of the presented cases underwent surgical separation on emergency or semi-emergency bases. Case 4 was operated upon at midnight because one twin expired from acute gastroenteritis and sepsis at the age of 44 days. Surgical separation was started within an hour after the death of the first twin. The separation of these omphalopaus twins took only one hour, but the second twin also expired an hour after the separation. The combined birth weight of this pair of conjoined twins was only 2,500 grams. In case 7, one twin had complex cardiac anomalies and con-

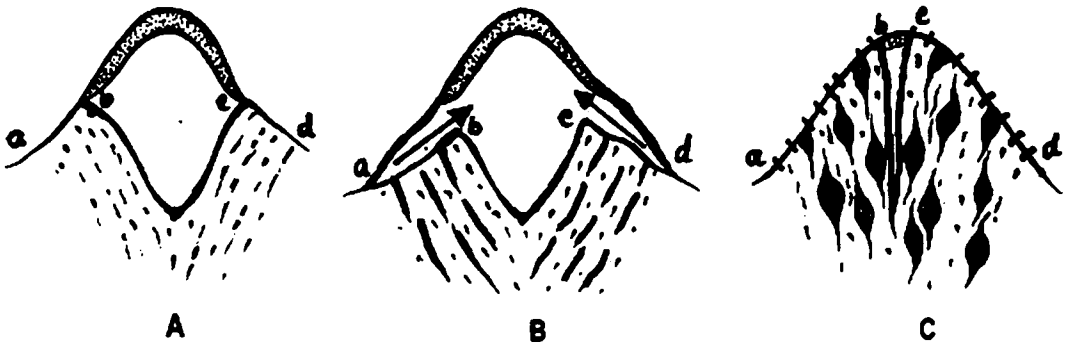

Fig. 10. Muscle advancement technique. A) A large gap between the medial boarder of the rectus muscles. B) The upper end of the rectus muscles is detached from the costal cartilage on each side, multiple longitudinal slits are made in the rectal sheaths and rectus muscles and the muscles are spread in horizontal direction to get a larger width. C) The upper end of the muscles are sutured to the costal margin.

gestive heart failure, which did not respond to medical treatment. Surgical separation was done on an semi-emergency basis, in the morning, at the age of 6 days. The twin with complex cardiac anomalies expired 18 days after surgery, while the other survived satisfactorily. Case 8 required an emergency procedure for the same reason. Surgical separation had to be done at 5.00 p.m. at the age of 29 hours. The surgical separation took only two hours. The twin with complex cardiac anomalies and intractable cardiac failure died on the operating table but the other survived satisfactorily.

Other indications for surgical separation in the newborn period may include the presence of other congenital anomalies or diseases that are not compatible with life without surgery but appear surgically correctable like intestinal obstruction, ruptured omphalocele or imperforate anus[30]. Unruptured omphalocele is not an indication for emergency separation, because this may be treated successfully with non-operative treatment. Even in twins with intestinal obstruction, it may be possible to do a temporary procedure in order to postpone a separation procedure until the patients are older. For instance, a colostomy or enterostomy may be temporarily done in cases of intestinal obstruction or imperforate anus[32]. Votteler [33] reported a pair of pygopagus conjoined twins, one of which developed necrotizing enterocolitis (NEC) with gut perforation at the age of 4 days. A colectomy and ileostomy were done for the twin, while the other did not receive general anesthesia. The twin recovered from NEC satisfactorily, and the surgical separation was postponed until 2 months of age.

Surgical separation of most omphalopagus and pygopagus conjoined twins appears to be less complicated than that of thoracopagus and ischiopagus. At our hospital, thoracopagus conjoined twins are considered inseparable if they have conjoined hearts at ventricular level. Ischiopagus separation is the most complex surgical procedure and requires a multidisciplinary approach of several surgical and medical specialties. Closure of defects after separation of conjoined twins with extensive fusion can be extremely difficult. It may be necessary to use synthetic prosthetic materials to temporarily cover the abdominal or chest wall defect[31]. Rotating skin flaps may be necessary in cases with insufficient skin and subcutaneous tissue. Pre operative planning should be carefully exercised. Techniques of pre operative pneumoperitoneum and tissue expanders have been reported to facilitate the closure[16,32,34-39]. Utilization of the skin and subcutaneous tissue of the fused third leg for soft tissue closure after separation of ischiopagus tripus twins has been reported[7,32,34,39].

After separation of the fusion area in thoracopagus conjoined twins, there is usually a big gap between the medial border of the rectus muscles. An attempt to suture the medial margin of the rectus muscles together to complete the peritoneal cavity closure is usually difficult in the upper portion,

because the medial portion of the costal margin is a "bare area" without muscular attachment (Fig. 10A). Muscle advancement technique may facilitate this closure in some cases (Fig. 10B, C)[11].

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ฝาแฝดตัวติดกัน : การผ่าตัดแยกในผู้ป่วย 11 คู่

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ผู้ป่วยฝาแฝดตัวติดกัน 11 คู่ได้รับการผ่าตัดแยกในสถาบันสุขภาพเด็กแห่งชาติมหาราชินี ผู้ป่วย 6 คู่มีการติดกันที่บริเวณลิ้นปี่ 4 คู่มีการติดกันของผนังทรวงอกและหน้าท้องและ 1 คู่มีการติดกันที่บริเวณก้นกบ ทารก 8 คู่เป็นเพศหญิงและ 3 คู่เป็นเพศชาย ผู้ป่วยฝาแฝด 3 คู่ จำเป็นต้องได้รับการผ่าตัดแยกกันแบบเร่งด่วนในระยะทารก ส่วนอีก 8 คู่ได้รับการผ่าตัดแยกกันโดยไม่เร่งด่วนเมื่ออายุมากขึ้น

ในกลุ่มที่จำเป็นต้องได้รับการผ่าตัดแยกเร่งด่วน ทารก 1 คู่ซึ่งมีน้ำหนักแรกคลอดรวมกันเพียง 2,500 กรัมจำเป็นจะต้องผ่าตัดแยกเพราะทารกคนหนึ่งเสียชีวิต จึงได้รับการผ่าตัดแยกภายใน 1 ชั่วโมงเพื่อพยายามจะรักษาชีวิตทารกอีกคนหนึ่ง อย่างไรก็ตามทารกคนหลังก็เสียชีวิต ภายในประมาณ 1 ชั่วโมงหลังผ่าตัด ฝาแฝดอีก 2 คู่ได้รับการผ่าตัดแยกฉุกเฉินเพราะคู่แฝดคนหนึ่งอาการเลวลงเนื่องจากมีความพิการโดยกำเนิดของหัวใจ ทารกที่มีความพิการของหัวใจเสียชีวิต หลังผ่าตัดหรือในขณะผ่าตัด แต่คู่แฝดของทั้ง 2 คู่รอดจากการผ่าตัดและเจริญเติบโตปกติ

ในกลุ่มผู้ป่วยที่ได้รับการผ่าตัดแยกแบบไม่เร่งด่วน 1 คู่มีฝาแฝดคนหนึ่งที่มีความพิการโดยกำเนิดของหัวใจชนิดแก้ไขลำบาก เมื่อฝาแฝดคู่นี้ได้รับการผ่าตัดแยกเมื่ออายุ 2 ปี 9 เดือน ฝาแฝดที่มีความพิการโดยกำเนิดของหัวใจเสียชีวิต 2 ชั่วโมงหลังผ่าตัด ส่วนคู่แฝดรอดจากการผ่าตัดแยกและเจริญเติบโตเป็นปกติ

ส่วนในผู้ป่วยอีก 7 คู่ที่ได้รับการผ่าตัดแยกแบบไม่เร่งด่วน ฝาแฝดทั้งสองคนของแต่ละคู่รอดจากการผ่าตัดแยก แต่ผู้ป่วยคนหนึ่งเสียชีวิต 10 วันหลังผ่าตัดอย่างไม่คาดหวัง สาเหตุการตายไม่ชัดเจน

คำสำคัญ : ฝาแฝดตัวติดกัน, ฝาแฝดสยาม, ฝาแฝดที่ตัวติดกันบริเวณหน้าอกและหน้าท้อง, ฝาแฝดตัวติดกันบริเวณลิ้นปี่, ฝาแฝด–ตัวติดกันบริเวณก้นกบ, การผ่าตัดแยก

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