

Maternal Referral Analysis On Conjoined Twin Pregnancy In Dr. Soetomo Hospital Surabaya During 2010-2019

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Abstract

Conjoined twin pregnancies are rare, and multidisciplinary approach in higher level of healthcare services are urgently needed. This paper aims to analyze the maternal referral system in the case of conjoined twin pregnancies in Dr. Soetomo Hospital to prevent delay of care for both mother and fetuses.

This cross-sectional study analyzed medical records from maternity ward in Dr. Soetomo Hospital from 2010 to 2019. The records were compiled then grouped based on several criteria as follows: gestational age at the time of referral, mode of delivery, referral source and neonatal outcomes.

Total referred cases of conjoined twin pregnancies are 16 cases with thoracoabdominophagus as the most common type of conjoined twins. 10 (63%) cases were delayed referral of ≥ 21 WGA. C-section was performed in 9 (56%) cases of delayed referral, and the remaining 6 (44%) cases by vaginal delivery. Neonatal outcomes are generally worse, 9 (55%) twins promptly deceased after delivered, 2 (13%) and 3 (19%) twins were deceased after 1 hour and 24 hours post partum respectively. Only 2 twins survived perinatal period but deceased 2 days later.

Early diagnosis and referral are mandatory for suspected conjoined twin pregnancies to reduce maternal and neonatal morbidities and mortalities. Congenital anomalies screening by ultrasound are highly encouraged in 11-13 WGA or at first trimester for early diagnosis.

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Keywords: Conjoined Twins; Referral; Congenital

1. Introduction

Conjoined twins are identical twins with conjoined or fused body parts in utero. This condition is a rare complication, and occurs in monochorion monoamniotic pregnancies. If the division occurs after the formation of the amnion, there will be 1 placenta, 1 chorion, 1 amnion so that a conjoined twin can occur or what we often call conjoined pregnancy (Slotnick, 2013).

The first successful separation of conjoined twins was carried out in Switzerland in 1689. The most famous cases of conjoined twins in the world are Eng and Chang from Thailand who were born in 1811 (Saguil, 2009). In Indonesia, there is no exact number of cases of conjoined twin pregnancy. At Dr. Soetomo Hospital, in 2009 the Center for Integrated Conjoined Twins Services (PPKST) was established at Dr. Soetomo Hospital of

Medical Faculty Airlangga University Surabaya. Records from 2010 to 2019 approximated the number of cases of conjoined twin pregnancies were 16 cases.

Every husband and wife yearn to have a healthy baby. This could be achieved through a normal pregnancy, where in a normal pregnancy the results of conception can continue to grow so that the baby born met these criteria. However, pregnancy can also be disrupted, causing pregnancy abnormalities. This disorder can be in the form of congenital abnormalities, conjoined pregnancy, and intrauterine fetal death. These pregnancy abnormalities depend on the stage and type of disorder. Conjoined twins pregnancy can be detected earlier using ultrasound at 12-13 weeks of gestation (McManara,2015).

This study will discuss the pregnancy profile of conjoined twins born in Dr. Soetomo Hospital Surabaya as well as efforts to implement a strategic approach to address delays in referrals, find out early diagnosis of conjoined twins, and reduce birth rates by C-section.

2. Research Elaborations

2.1 Methods

This study was an observational analytic research using cross sectional design to compile medical records in Dr. Soetomo Hospital during 2010-2019. Data collection was using secondary data in the form of Medical Records (RM) which were then processed using Microsoft Office Excel. The location of the research was carried out at Dr. Soetomo Hospital Surabaya. The data collection and processing was carried out from 2010 to 2019. The sample is part of the population that has met the inclusion criteria, namely patients with conjoined twin pregnancies who were referred to the Pregnancy Outpatient Clinic and treated in the Obstetric Ward of Dr. Soetomo Hospital Surabaya from 2010-2019.

3. Result

3.1 Case Distribution of Conjoined Twin Pregnancies

From the results of the data collection on the Obstetric Outpatient Clinic register book and delivery room at Dr. Soetomo Hospital in the period January 2010 - December 2019, there were 16 referral cases with conjoined twins. During 2018 there were the most conjoined twin pregnancy referral cases, there were 4 cases recorded in 2018.

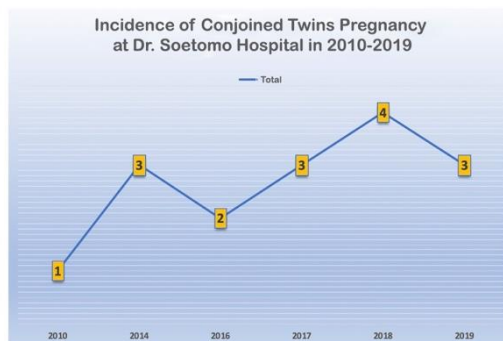


Figure 1. Incidence of Conjoined Twin Pregnancies in Dr. Soetomo Hospital during 2010-2019

Based on the data, 75% cases of conjoined twins were Thoracoabdominophagus (12 patients), 13% were Paraphagus (2 patients), 6% were Cephalophagus (1 patient) and the remaining 6% were Omphalophagus (1 patient) from a total of 16 samples of conjoined twin pregnancies in Dr. Soetomo Hospital Surabaya which met the inclusion and exclusion criteria.

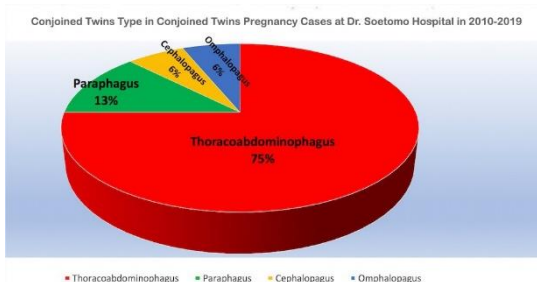


Figure 2. Type of Conjoined Twins in Dr. Soetomo Hospital 2010-2019

3.2 Characteristics of Late Referral Cases of Patients with Conjoined Twin Pregnancy

From the data of this study, the number of pregnant patients with conjoined twins were 16 cases. Of these 16 cases, 10 cases were late referrals and 6 cases were on time referrals. Of the 10 late referral cases, 9 of these patients were scheduled for termination by C-section.

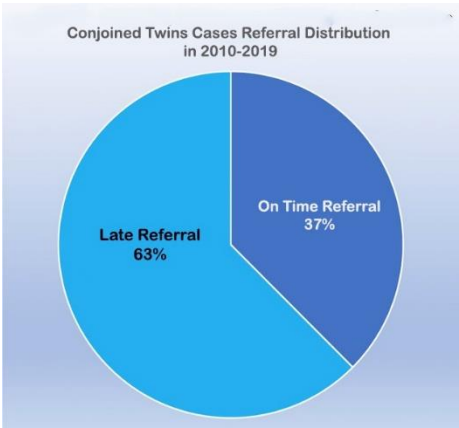


Figure 3. Referral Distribution

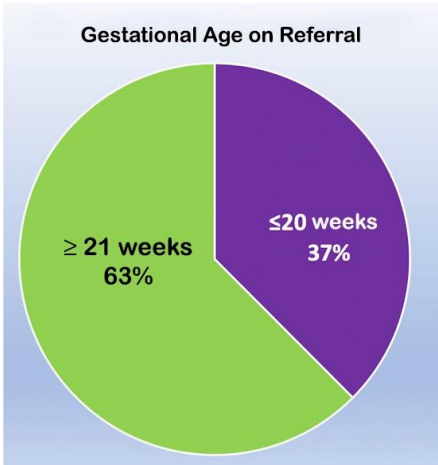


Figure 4. Gestational Age on referral

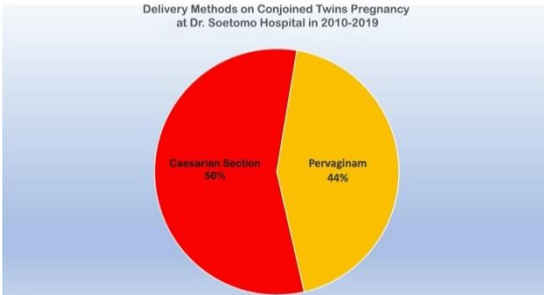


Figure 5. Delivery Method

3.3 Patient Characteristics of Conjoined Twin Pregnancy Cases based on Risk Factors

From the data tracing, it was found that the average age of the patients referred to Dr. Soetomo Hospital Surabaya for the period January 2010 - December 2019 is 28 years. The youngest was 22 years old, while the oldest was 40 years old. Based on the age range group, the proportion in the 20-25 year age range group was 31%, in the 26-30 year age range group the proportion of conjoined twins was 44% and in the 31 - 34 year age group it was 6%. Whereas in the age group ≥ 35 years, the proportion of conjoined twins was 19%.

Table 1. Maternal Age Distribution in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Age	Total (%)
20-25 years old	5 (31%)
26-30 years old	7 (44%)
31-34 years old	1 (6%)
≥ 35 years old	3 (19%)

Table 2. Maternal Parity Distribution in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Parity	Total (%)
Primigravida	8 (50%)
Multigravida	8 (50%)

Table 3. Distribution of Maternal Marital Status in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Marital Status	Total (%)
Married 1x	12 (75%)
Married 2x	1 (6%)
Married 3x	1 (6%)
Not married	2 (13%)

3.4 Characteristics of Late Diagnosis and Referral of Patients with Conjoined Twin Pregnancy Based on Complications when Referred to Dr. Soetomo Hospital Surabaya

In this case report, the co-morbidities of conjoined twins by obesity accounted for 31% of sample, followed by advanced maternal age ≥ 35 years in 19% of sample.

Table 4. Maternal Comorbidities in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Comorbid	Total (%)
Gestational Diabetes	1 (6%)
Severe Preeclampsia	2 (13%)
Heart Disease	1 (6%)
Obesity	5 (31%)
Anemia	1 (6%)
Chronic Hypertension	1 (6%)
Age ≥ 35 years old	3 (19%)
No comorbid	2 (13%)

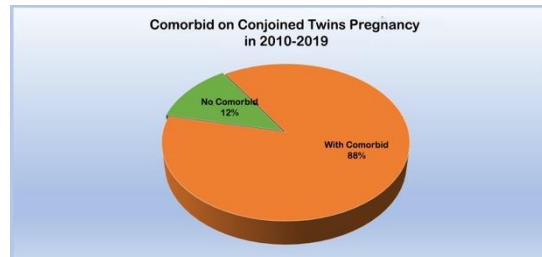


Figure 6. Comorbid factor of Conjoined Twins

3.5 Neonatal outcome in Conjoined Twin Pregnancy Cases

Patients with conjoined pregnancies have poor prognosis towards neonatal outcomes. The most common outcomes of conjoined twins are infant mortality. The highest number of babies died at birth as much as 55%, then 19% of the babies who could survive up to the age of 1 day. The longest survival rate for infants was 29 days.

Table 5. Characteristics of Neonatal Outcomes in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Baby Outcome	Total (%)
Immediate death	9 (55%)
30 minutes – 1 hour	2 (13%)
> 1 hour – 24 hours	3 (19%)
≥ 24 hours – 30 days	2 (13%)

3.6 Late Referral Patterns in Conjoined Twins Pregnancy Cases

From the 16 cases referred as conjoined pregnancy cases, 7 cases were referred at gestational age <25 weeks and 9 cases were referred at later gestational age ≥ 25 weeks. 13 cases were referred from outside Surabaya and 3 cases were from Surabaya. Most of the referrals from outside Surabaya came from Solo, Madiun, Sidoarjo, with each 2 cases. While the others came from Gresik, Mojokerto, Banyuwangi, Sorong, Nganjuk, Malang, Sampit for 1 referral case each.

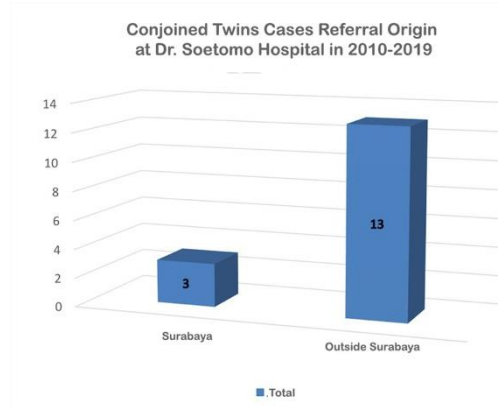


Figure 7. Referral Origin

Table 6. Referral Origin of Conjoined Twin Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Referral Origin	Total (%)
Surabaya	3 (19%)
Sidoarjo	2 (13%)
Madiun	2 (13%)
Solo	2 (13%)
Gresik	1 (7%)
Mojokerto	1 (7%)
Banyuwangi	1 (7%)
Malang	1 (7%)
Nganjuk	1 (7%)
Sampit	1 (7%)
Sorong	1 (7%)

Table 7. Mode of Delivery in Conjoined Twins Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Delivery Method	Total (%)
Pervaginam	7 (44%)
Caesarean Section	9 (56%)

4. Discussion

The number of cases of conjoined pregnancy in the world is actually unknown. However, it is estimated that the prevalence of conjoined twins in the world is 1: 100,000 births. In Indonesia, there is no official record regarding the number of cases of conjoined pregnancy prevalence. But interestingly, from the results of observations at Dr. Soetomo Hospital Surabaya during the period January 2010 - December 2019 there were 16 cases of conjoined twin pregnancy which were neatly recorded in medical record status.

Epidemiological data are limited due to the rarity of the disease and also the inaccuracy of data recording and keeping regarding the number of pregnancies in the population. A number of arguments suggest that the variation between regions is due to problems in reporting, socioeconomic factors and nutritional factors. Problems with reliable reporting and epidemiology can be caused by a number of factors such as population-based data reporting compared to hospital-based data, inconsistencies in defining cases, inability to determine population at risk, absence of a reliable data center. centralized, as well as poor selection of control groups to compare risk factors that may contribute. The incidence is very difficult to calculate because not all cases will be reported or recognized, at this time there is no data center that records the number of pregnancies in the Indonesian population (Handriani, 2015).

From the data compiled, there were 10 cases out of 16 cases of conjoined twin pregnancy with late referral at Dr. Soetomo Hospital Surabaya during the period January 2010 - December 2019 that already knew that they had a diagnosis of conjoined twin pregnancy. Whereas, on time referral to a tertiary hospital before 25 weeks of gestation made the pregnancy able to be delivered pervaginam, thereby reducing the number of C-Section where the C-Section procedure itself can leave defects in the mother's uterus.

Not all level II regional hospitals have subspecialist obstetricians and are capable of providing essential obstetric services with basic essential obstetric and comprehensive obstetric services. The problem in managing conjoined twins is the diagnosis that must be enforced by an obstetrician specialist and there must also be a good and sophisticated ultrasound devices. If the obstetrician can establish a diagnosis of conjoined twin

pregnancy as early as possible by ultrasound, then the patient can be terminated vaginally so that it will reduce the delivery of C-Section.

During the period of January 2010 - December 2019, there were 10 late referral cases out of 16 cases of conjoined twin pregnancy. These 10 late referral cases were lately diagnosed in the previous hospital, so when they came to Dr. Soetomo Hospital, their pregnancy has already entered the 3rd trimester, so that many of these cases have had to be delivered by C-Section. From the 10 late referral cases, 9 cases were terminated by C-Section. The late referral analysis is listed in the following table.

Table 8. Distribution of Referrals in Conjoined Twin Pregnancy Cases at Dr. Soetomo Hospital Surabaya

Referral Distribution	Total (%)
On Time Referral	10 (63%)
Late Referral	6 (37%)

Table 9. Gestational Age When Referred to Conjoined Twins Pregnancy Case at Dr. Soetomo Hospital Surabaya

Referral Distribution	Total (%)
14/15 weeks	1 (6%)
16/17 weeks	1 (6%)
17/18 weeks	1 (6%)
21/22 weeks	1 (6%)
22/23 weeks	1 (6%)
23/24 weeks	1 (6%)
25/26 weeks	5 (33%)
29/30 weeks	1 (6%)
32/33 weeks	3 (19%)
36/37 weeks	1 (6%)

There is no literature that determines the exact time of delivery in cases of conjoined pregnancies. However, several case reports recommend termination at 35 weeks of gestation after corticosteroid administration to avoid the risk of IUFD and prematurity. From several case reports, termination with C-Section was performed to avoid umbilical cord entanglement (Van Mieghem, 2014).

Vaginal deliveries in previously undiagnosed conjoined twins are at high risk for shoulder dystocia, uterine rupture and IUFD. Vaginal delivery in conjoined twins can be carried out in the 2nd trimester of gestation where the baby's size is smaller than at term, and is carried out in conjoined pregnancies with babies who have a small chance of survival after birth (Harma, 2005).

In 16 cases of conjoined pregnancy in Dr. Soetomo Hospital, the most common method of delivery was by C-Section (9 cases). As many as 7 cases can be diagnosed by fetomaternal ultrasound at ≤ 20 weeks of gestation, so that they can be delivered vaginally. Before deciding on termination, the Fetomaternal team at Dr. Soetomo Hospital conducted multidisciplinary meetings with other departments. From the 16 cases of conjoined pregnancy in Dr. Soetomo, 11 cases have gone through the case conference first.

Table 10. Case Conference of Conjoined Twins Pregnancy cases in Dr. Soetomo Hospital Surabaya

Case Conference	Total (%)
Discussed in Case Conference	11 (67%)
Not discussed	5 (33%)

5. Conclusion

There were 16 referral cases of conjoined twin pregnancies at Dr. Soetomo Hospital Surabaya during Januari 2010-December 2019. From the 16 referral cases, 10 cases were late referral cases (62.5%), 14 cases were with maternal comorbidities (88%), 13 cases were referred from outside Surabaya and 3 cases were referred from Surabaya. All of these late referral cases were referred as non-emergency cases. Patient characteristics are the same between primigravida and multigravida, with the most common comorbid was obesity. Most patients were referred during the 3rd trimester of pregnancy, as many as 7 patients. All cases of conjoined twin pregnancies referred to Dr. Soetomo Hospital must have done re-diagnosing by fetomaternal team, then planned for the termination of the pregnancy. From 16 cases, 9 patients underwent termination with C-Section and 7 patients underwent pervaginam termination.

Ethical Approval

The research was approved by the Human Research Ethics Committee, Dr. Soetomo Hospital, approval number 0392/LOP/301.4.2/III/2021.

Conflicts of Interest

The authors declare that there are no conflicts of interest.

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