

Conjoined twins and legal authorization for abortion

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SUMMARY

Objective: To describe pregnancies with conjoined twins according to the request for legal termination of pregnancy. **Methods:** Retrospective review of pregnancies with conjoined twins, with no possibility of extrauterine survival or postnatal surgical separation, observed at a tertiary teaching hospital, between 1998 and 2010. **Results:** Amongst 30 cases seen during the study period, six (20.0%) couples decided to continue with the pregnancy, termination of pregnancy was not requested due to advanced gestational age (> 25weeks) in 5 cases (16.7%). Legal authorization to terminate the pregnancy was requested in 19 (63.3%) cases: permission was granted in 12 (63.2%), denied in five (26.3%) and information was missing in two (10.5%) cases. A successful vaginal delivery was performed in 83.3% of the cases undergoing termination of pregnancy and a cesarean section was performed in all the remaining cases ($p < 0.01$). **Conclusion:** In pregnancies with conjoined twins and without fetal prognosis, legal termination of the pregnancy is an alternative. Moreover, a successful vaginal delivery can be performed in most cases before the third trimester, further reducing maternal risks and parental suffering.

Keywords: Twins conjoined; legal abortion; ultrasonography prenatal; pregnancy complications; pregnancy multiple.

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INTRODUCTION

In the last decades, technological advances in Reproductive Medicine have been associated with the increase in multiple pregnancies, as well as the complications associated with this event and among them, the occurrence of fetal abnormalities. Among the fetal development abnormalities, the most severe one is the development of conjoined or Siamese twins, with an estimated incidence of 1:50,000 to 1:200,000 gestations¹⁻³.

Conjoined twins are classified according to the most prominent site of connection, together with the term *pagus*, which indicates fusion: thoracopagus (thoracic union); omphalopagus (abdominal union); parapagus (extensive lateral union); pygopagus (sacral union); ischiopagus (lower abdomen and pelvis union); cephalopagus (head and encephalon union); craniopagus (cranial union) and rachipagus (vertebral column union)⁴.

The ultrasonographic diagnosis can be carried out from the 12th week of gestation⁵ and the early detection is crucial for planning the conduct in the pregnancy, as well as for family counseling⁶. Survival in cases submitted to surgical separation depends on the degree of organ sharing between the fetuses and associated malformations. In this context, cardiac unions are the conditions associated with worse prognosis^{7,8}. Many conjoined twins, without perspective of postnatal surgical separation, die within a few hours after birth⁹.

In Brazil, the legal permission for abortion includes only the situations where the pregnancy was the result of rape or when there is risk of death for the mother. In cases of lethal fetal malformation, such as anencephaly, the couple can apply for legal permission to terminate the pregnancy. As it is a rare event, few reference centers have cases of conjoined twins whose parents applied for legal authorization to terminate the pregnancy.

The Judicial System is also rarely called upon to decide on these cases of such elevated complexity. In cases where fetal survival has been considered impossible, the abortion has been authorized, which abbreviates the mother's suffering and minimizes the risk of complications during the pregnancy and birth.

When there is extensive vital organ-sharing, in which the postnatal separation is not possible and when that is the parent's wish, the legal authorization for the abortion has been requested. The objective of the present article was to describe cases of conjoined twins in whom the postnatal surgical separation was not possible due to severe organ-sharing and extrauterine survival was considered impossible, describing the cases according to the request or not for legal authorization to terminate the pregnancy.

METHODS

Cases of multiple pregnancies with a diagnosis of conjoined twins followed by the Service of Multiple Pregnancy of a university hospital, from June 1998 to June 2010 were ret-

rospectively analyzed. The study was approved by the Ethics Committee in Research of the institution, protocol # 0212/10.

The following inclusion criteria were adopted: diagnosis of twin pregnancy with conjoined twins at the first assessment in this service; conclusive assessment of postnatal surgical separation or survival and follow-up at the institution. The data were collected from the medical files and hospital birth registers.

A total of 37 cases of pregnancies with conjoined twins were identified. One case had a diagnosis of fetal death at the first ultrasonographic assessment and was not included in the analysis. In six cases there was a prognosis of possible postnatal surgical separation and were not included in the analysis, either. The present study analyzed the data from 30 pregnancies with a diagnosis of conjoined twins in whom postnatal surgical separation or survival was considered impossible. The data concerning the characteristics of this population are shown in Table 1.

The pregnant women were evaluated for more than one ultrasonographer from the Fetal Medicine team, to characterize the site of twin union and evaluate the extension of organ-sharing. The presence of other associated malformations was also investigated and a specialized fetal echocardiography was performed to determine prognosis and the possibility of surgical separation when the fetuses shared the heart.

The following data were investigated: gestational age at diagnosis, prognosis related to fetal lethality, legal authorization or not to terminate the pregnancy, type of delivery, birth complications, product of conception weight, gestational age and the following demographic variables: maternal age, marital status, parity, number of living children and prior cesarean section (C-section). The weight of the conjoined twins was measured in the birth room.

The following criteria were adopted for requesting legal authorization to terminate the pregnancy: lethality of the fetal malformation and fetal union; lack of prognosis for postnatal surgical separation; gestational age < 25 weeks and no contra-indication for vaginal delivery. In cases of gestational age > 25 weeks, or when the couple did not want to terminate the gestation, the pregnancy was followed until its resolution.

In these cases, the parents received multidisciplinary assistance on offers of support measures to guarantee the basic comforts to the twins after the birth.

In cases where the couple chose to request legal authorization to terminate the pregnancy, the ultrasonography and echocardiography reports were provided, which included details of the examination findings and prognostic report on the fetuses.

The pregnant woman/couple was submitted to psychological assessment that demonstrated that was the "adequate" decision and a specific report was written on the

Table 1 – Maternal characteristics in cases of conjoined twins and type of fetal union according to the request or not of legal authorization for pregnancy termination

	Requested termination n = 19	Did not request termination n = 11	p
Age, years, median (min-max)	26 (19-40)	24 (22-42)	0.310
Gestational age at diagnosis, weeks, median (min-max)	20 (14-31)	27 (24-32)	< 0.001
Ethnicity			
Caucasian	10 (52.6%)	6 (54.5%)	0.781
Non-caucasian	9 (47.4%)	5 (45.5%)	
Parity			
Nulliparous	10 (52.6%)	3 (27.3%)	0.260
≥ 1	9 (47.4%)	8 (72.7%)	
Living children			
Yes	8 (42.1%)	7 (63.6%)	0.449
No	11 (57.9%)	4 (36.4%)	
Previous C-section			
Yes	0 (0%)	2 (18.2%)	0.126
No	19 (100%)	9 (81.8%)	
Type of union			
Thoracopagus	16 (84.2%)	7 (63.6%)	0.277
Parapagus dicephalus	2 (10.5%)	3 (27.3%)	
Cephalopagus	0 (0%)	1 (9.1%)	
Omphaloischiopagus	1 (5.3%)	0 (0%)	

diagnostic and prognostic findings and psychological assessment. The report was signed by two doctors from the Fetal Medicine team that participated in the assessment. Copies of literature articles on the fetal condition, supporting the lethal condition of the case, were added to the report. The pregnant woman/couple was advised to present the legal authorization request to terminate the pregnancy at a Court of Justice in the city/town where they lived. After the judge's decision, the patient returned to the service to follow the decision.

The prognosis of lethality was characterized by extensive union of vital organs (heart and liver), with complex cardiac malformation. Once the legal authorization was granted to terminate the pregnancy, fetal death was induced, followed by vaginal delivery. Cervix preparation and birth induction were initiated on the same day after fetal death was induced, when the ultrasonographic assessment showed that the largest diameter of the conjoined parts and the fused cephalic poles would not be an impediment to birth.

The results were analyzed using the software Statistica for Windows (release 4.3, Statsoft, Inc., 1993). The variables were analyzed descriptively, by calculating medians, minimum and maximum values, absolute and relative frequencies. The categorical and semi-quantitative data were assessed by the Chi-square test or Fisher's Exact test, when

indicated. Continuous variables were analyzed by Mann-Whitney U test. The level of significance was set at $p < 0.05$.

RESULTS

A total of 30 cases of multiple gestations with conjoined twins were assessed, with a prognosis of impossible post-natal surgical separation or survival. Of the 25 cases to whom the possibility of requesting legal authorization to terminate the pregnancy was offered, 76% chose to terminate and 24% (six cases) chose to maintain the pregnancy. The characteristics of the gestations in the cases that requested or not legal authorization to terminate the pregnancy are shown in Table 1. In five cases with gestational age > 25 weeks at the diagnosis of conjoined twin pregnancy, the termination was not discussed. Of the 19 cases for which the legal authorization to terminate the pregnancy was requested, 12 (63.2%) requests were granted by the judge and the abortion was authorized, five (26.3%) requests were denied and two (10.5%) did not return to our service and were lost to follow-up. The characteristics of the 17 cases for which the legal decision to terminate or not was known are shown in Table 2. In the group that chose to terminate the pregnancy, vaginal delivery occurred in 83.3%, but in the group for which the legal authorization was denied, a C-section was performed in 100% of the cases ($p < 0.01$).

Table 2 – Characteristics of the diagnoses and birth data in cases of conjoined twins according to the legal authorization or not to interrupt the pregnancy

	Authorized n = 12	Non-authorized n = 5	p value
GA at diagnosis, weeks, median (min-max)	20 (14-32)	24 (22-26)	0.050
Type of union			
Thoracopagus	10 (83.3%)	5 (100%)	1.00
Parapagus dicephalus	2 (16.7%)	0 (0%)	
GA at abortion/delivery, weeks, median (min-max)	22 (19-33)	34 (33-38)	0.002
Type of birth			
Vaginal	10 (83.3%)	0 (0%)	0.003
C-Section	2 (16.7%)	5 (100%)	
Weight of twins, g, median (min-max)	693 (370-2850)	4270 (3660-4700)	0.009

GA, gestational age.

Table 3 shows the description of the cases for which the legal authorization to terminate the pregnancy was granted. The five cases that were denied authorization are shown in Table 4. Three cases had other associated malformations, in addition to the cardiac ones. As for the cardiac union, all cases had atrial and ventricular union and in four cases the twins died within the first three hours after birth and in one case, on the 28th day after the birth.

DISCUSSION

Although there have been many publications on conjoined twins, few have analyzed the characteristics of the cases in which the pregnancy was terminated. In countries where abortion is legal, couples are free to choose this option. However, in Brazil, as the presence of fetal anomalies incompatible with extrauterine survival is not among the situations established by the Law as the ones that allow the termination of pregnancy (when the preg-

Table 3 – Clinical description of cases of conjoined twins in which the mother received legal authorization to terminate the pregnancy

Case	Type of union	Shared organs	Associated malformations	GADx	GAI	Type of delivery	Weight (g)
6	Thoracopagus	Heart, liver	Cardiac	19w4d	25w4d	Vaginal	1.510
7	Thoracopagus dibrachius tripus	Heart, liver	Cardiac, hygroma, abdominal wall	20w2d	21w2d	Vaginal	700
8	Thoracopagus	Heart, liver	Cardiac	31w4d	32w4d	C-section	2.850
9	Thoracopagus	Heart, liver	Cardiac	16w1d	19w6d	Vaginal	560
10	Parapagus dibrachius dipus	Heart, liver, intestines, urinary system	Cardiac, F1 with diaphragmatic hernia, extremities	14w3d	18w3d	Vaginal	370
12	Thoracopagus tripus	Heart, liver	Cardiac, omphalocele and extremities	24w4d	27w4d	C-section	1.040
25	Thoracopagus	Heart, liver	Cardiac	21w3d	25w2d	Vaginal	1.080
26	Thoracopagus	Heart, liver	Cardiac	25w4d	27w	Vaginal	1.430
28	Thoracopagus	Heart, liver,	Cardiac F1 and F2 with cleft lip and palate	18s	21w2d	Vaginal	685
30	Thoracopagus	Heart, liver	Cardiac; F1 with cystic hygroma; F2 with spina bifida	19w4d	22w	Vaginal	495
36	Thoracopagus	Heart, liver	Cardiac; F1 with cleft lip and palate	17w5d	19w4d	Vaginal	561
20	Parapagus dicephalus tribrachius dipus	Heart, liver, intestine	Cardiac	18w	21w3d	Vaginal	530

GADx, gestational age at diagnosis; GAI, gestational age at interruption; F1, fetus1; F2, fetus 2.

Table 4 – Clinical description of cases of conjoined twins in which the mother was denied legal authorization for interrupting the pregnancy

Case	Type of union	Organs shared	Associated malformations	GAD weeks	Weight (g)	Apgar NB1 1st/5 th min	Apgar NB2 1st/5 th min	OTI	Age death
1	Thoracopagus	Heart, liver	Cardiac, gastroschisis, F2 cleft lip and palate	35w5d	4270	2 / 5	2 / 5	Yes	3h
2	Thoracopagus	Heart, liver	Cardiac, F1 diaphragmatic hernia, F2 cleft lip and palate	33w4d	–	4 / 4	4 / 4	No	2h
14	Thoracopagus	Heart, liver	Cardiac, omphalocele	37w6d	3660	6 / 3	6 / 3	No	1h
28	Thoracopagus	Heart, liver	Cardiac	34w2d	4700	3 / 4	3 / 4	Yes	28d
33	Thoracopagus	Heart, liver	Cardiac	33w	–	–	–	–	1d

GAD, gestational age at delivery; F1, fetus 1; F2, fetus 2; NB, newborn; OTI, orotracheal intubation.

nancy was the result of rape or when there is risk of death for the mother), couples who decide to interrupt the pregnancy must request legal authorization to undergo the procedure, prolonging parental suffering and anxiety.

The present study demonstrated that, when the malformation lethality was confirmed, 76% of the couples chose to request legal authorization to terminate the pregnancy. Of the cases for which the authorization was requested, 63% obtained a favorable decision and the abortion was authorized. The median gestational age at pregnancy interruption was 22 weeks. The mean interval to obtain the legal authorization was approximately three weeks in the 12 cases that had a favorable decision. In cases where the Justice System denied authorization to terminate the pregnancy, the delivery was carried out by C-section and all twins died, four cases on the same day of the birth and one case on the 28th day after the birth.

Pregnancy termination in cases of conjoined twins and no possibility of extrauterine survival aims at minimizing the maternal risks associated with the difficulties posed by the birth of conjoined fetuses, which are invariably delivered by C-section when they reach the third gestational trimester, as well as the risks inherent to the maintenance of a twin pregnancy. The abortion performed up to the second trimester allows a vaginal birth, reducing the risk of maternal complications and without submitting the woman to a procedure that can damage her uterus and her reproductive health, mainly when the uterine incision is in a corporal (vertical) portion. Once submitted to a corporal C-section, the subsequent births will mandatorily occur by C-section, as the risk of uterine rupture is well-known in pregnant women with a previous history of corporal C-section.

The Court of Justice of the State of São Paulo, in a decision regarding the authorization to terminate a pregnancy with conjoined twins, affirms: “*This Court of Justice has decided that, considering the impossibility of extra-uterine survival, the abortion shall be permitted, with the objective of abbreviating maternal suffering, out of deference to their grief, their health and dignity*” (Legal authorization # 990.09 100287-9 - Santos – 6th Court of Justice - Judge Marco Antonio - 21.05.09).

The impossibility to perform the postnatal surgical separation of conjoined twins is determined by the severity and complexity of cardiac union and in cases that can result in unacceptable severe deformities. This situation includes thoracopagus twins with complex cardiac union and the cephalopagus or craniopagus, when the surgical separation can result in neurological sequelae^{10,11}. Throughout 12 years, 37 twin pregnancies with conjoined twins were diagnosed in this institution. That is a relevant series of cases, when compared to the ones described in the literature¹²⁻¹⁷. In an epidemiological study carried out in Spain², the authors analyzed the incidence of conjoined twins in two periods (1980 to 1985 and 1986 to 2006) and reported a decrease in the incidence of conjoined twin births after the abortion legalization.

The median gestational age at the first ultrasonographic assessment was relatively late in the second trimester, 24 weeks, when compared with two series in the USA published in 1990¹⁸ and 2002¹⁴, in which the mean gestational ages were 18.5 and 19.8 weeks and in which 64.3% and 21.4% of the cases the pregnancy was terminated in the first weeks of the second trimester.

In a literature review of cases of conjoined twins diagnosed below the 15th week, Pajkrk and Jauniaux¹⁹ found 50 published cases, of which 41 (82%) decided to terminate the pregnancy.

The more frequent indication for routine ultrasonography in the first trimester of pregnancy in several countries provides an early diagnosis of fetal abnormalities. The correct diagnosis, before the viability, allows the parents to choose to request or not legal authorization for pregnancy termination, and, consequently, minimizes maternal morbidity, as the pregnancy interruption at an earlier gestational age has fewer risks. In this study, in the cases in which the authorization was granted and the abortion was performed, the necropsy assessment confirmed the findings described in the prenatal assessment and all cases in which the pregnancy was maintained resulted in neonatal death without surgical separation.

In the present study, the request for legal authorization for pregnancy termination followed the recommendations of the Regional Council of Medicine of the State of São Paulo (*Conselho Regional de Medicina do Estado de São Paulo*)²⁰. The gestational age at the time of pregnancy termination is clinically important, as the earlier the abortion is performed, the lower the maternal risks are. Vaginal delivery is successful due to the flexibility of soft tissue connecting the fetuses and due to the smaller total volume of fetuses.

Although the literature has many reports on conjoined twins, addressing several medical, ethical and legal aspects, maternal complications are seldom described and detailed. In a review of cases performed by Harper *et al.*²¹, comprehending published cases of omphalopagus in three hundred years, the authors found a description of dystocia in 36% of them and a stillbirth rate of 19%. Rode *et al.*¹⁶ described a 40-year experience with cases of conjoined twins and reported one maternal death during delivery. In the series described by Al Rabeeah¹² birth canal trauma is reported in two of five cases that underwent vaginal delivery.

Conjoined twins attract many ethical and legal arguments²²⁻²⁵. With the advances in Medicine, several diagnostic evaluations allow detailed assessments of their anatomical features. The ethical and medical challenges appear when the twins share vital organs, such as the heart and liver, preventing postnatal surgical separation or extrauterine survival. This vital organ-sharing can accompany fetal losses, neonatal death and early deterioration of one twin.

In practice, in cases of conjoined gestations where extrauterine survival is not possible, the physicians must inform the parents clearly, respecting their opinions, their cultural factors and religious beliefs. The decision must be made by the parents, after being counseled by the medical team, considering the best interest of the pregnant woman's health. From the prenatal diagnosis, during the pregnancy follow-up, the parents must discuss and plan with the medical team the possible measures to be adopted. Confidentiality must be guaranteed by all involved and public and media sensationalism must be avoided at

all costs^{23,25}. The approach in each case must be coherent with the decisions made jointly and the parents must be encouraged to seek religious counseling, if they wish so. Moreover, the parents must be assured that they have total control over the decision process regarding the conducts to be adopted.

Conjoined twin pregnancy represents a high-severity diagnosis, as in many cases the shared organs and the extension of fetal union result in characteristics that prevent postnatal surgical separation or extrauterine survival.

The choice to terminate the pregnancy can be attained by requesting legal authorization for the abortion. Once the request is granted in the second trimester of pregnancy, vaginal delivery is the usual procedure, decreasing the risks to the pregnant woman's health and minimizing parental suffering.

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