



A Case Report and Review of the Literature on Monomphalic Thoraco-Omphalopagus Conjoined Twins in the Congolese Environment

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Abstract

Conjoined twins are a form of the embryonic developmental anomaly of twin pregnancy of which there are several forms in the medical literature. The anomaly of embryogenesis, which has not yet been elucidated, is characterized by the fusion or fission of the embryonic disc, resulting in the fusion of the twins and the sharing of one or more organs. The origin is not mystical or religious, which is important for health care personnel to know, as they are called upon to assist the parents and their families (victims) with advice and clarification. We report the conjoined thoraco-omphalopagus twins recorded in a rural setting, 40 years old and multiparous woman, without any morbid history, referred for prolonged labour from a health facility to the general referral hospital of Lubao, in the province of Lomami, in the Democratic Republic of the Congo. It appears to be the first to be published in our setting, the aim of which is to present the characteristics of conjoined twins and their evolution in the context of our setting.

Subject Areas

Nursing

Keywords

Siamese, Conjoined Twins, Lubao, Lomami, DRC

1. Introduction

Conjoined twins, more commonly known as Siamese twins (or even teratogenic

twins), are a serious embryonic developmental anomaly in monochorionic twin pregnancies [1] [2] [3]. Their incidence in the literature is one case in ten to two hundred thousand live births and about one per cent of affected monozygotic twin pregnancies [1] [4] [5] [6]. Some authors suggest an incidence of one case in 250,000 including stillbirths [5]. All countries are concerned. Several forms of conjoined twins have been described, related to the arrangement of the different parts of the body: thoracopagus, omphalopagus, pyropagus, ischiopagus, cephalopagus, craniopagus, caudal parapagus, fetus-in-fetus and parasitic twins [3] [6] [7] [8].

Despite the technical and technological advances recorded in recent years in anaesthesia, resuscitation, surgery, and medical imaging, the prognosis remains poor because of the risk of mortality, especially in the case of the most severe forms such as craniopagus conjoined twins [6] [9] [10]. The treatment, when all the conditions are met, is multidisciplinary, because of the technical, psychological, ethical, and philosophical aspects [6] [11]. In particular, the treatment is surgical and consists of the separation of two twins [10] [12]. In certain circumstances, such as dichorionic diamniotic pregnancies [6] and teratology incompatible with ectopic life, it may be necessary to terminate the pregnancy [6] [8]. Hence the importance of ultrasound examinations or magnetic resonance imaging in a medical environment to finally ensure the antenatal diagnosis of conjoined twins [6].

In the scientific literature, the majority of described cases cover the European, Asian and American continents [7]. African cases are almost not described. In the Democratic Republic of the Congo, where there is no registry of congenital malformations, conjoined twins have been reported in newspapers for the general public, but not in the medical literature. This article reports the case of conjoined twins born at the general referral hospital in Lubao, Democratic Republic of the Congo. This description seems, in our opinion, the first to be published. It aims to present the characteristics (anatomical, diagnostic and prognostic) of the reported conjoined twins and their evolution in the neonatal period.

2. Patient and Observation

The patient for this study was Mrs. MM, a 40-year-old teacher living in Twendeleye, in the administrative division of Lubao, Lomami province, Democratic Republic of the Congo. The parturient obstetric formula was P₆G₆A₀D₀ (= parity 6, gravidity 6, zero abortion, and death) [five living children] and the date of last menstrual period was unknown. Mrs. MM. was referred from “la Prouesse health post” to Lubao General Referral Hospital on June 09, 2022 [12:02 p.m. local time: Universal Time +2] for prolonged labour with rupture of the water bag (amniotic fluid) for ten hours. In her history, she had followed four times prenatal consultations and had no history of congenital malformations in the family, blood pressure, or diabetes mellitus. Mrs. MM. lives in a monogamous marriage (no relevant information was obtained from the husband, due to lack of

informed consent). On clinical examination, uterine height was 36 Centimeters, fetal heart sounds (BCF) were present (127 beats per minute), active fetal movements (AFM) were present, cervix dilated to 4 Centimeters, and effaced to 25 per cent. The presentation was cephalic and the fetal attitude was undetermined. Given this picture, an emergency cesarean section was indicated. The preoperative workup included the determination of haemoglobin level [10 g% with the haemoglobinometer technique], blood grouping [O, positive rhesus], bleeding time [5 minutes], and clotting time [2 minutes]. A stable venous approach was set up with saline while waiting for the search for the donor of iso-group, iso-rhesus, and minor blood group compatible blood. The cesarean section was performed under local anaesthesia by infiltration (with Lidocaine) and then general anaesthesia (Ketamine at the dose of 0.5 mg/Kg) during the hysterotomy phase. The surgical team was led by the general practitioner, and the anaesthetic team was led by a nurse not specialized in anaesthesia and resuscitation. The duration of the cesarean section was thirty-eight minutes starting at 13:40' and ending at 14:18'. The two extracted conjoined twins did not cry at extraction and had a pink colouration [Figure 1], with vernix caseosa. Gestational age was estimated (based on the appearance of the newborns) between 34 and 36 weeks of amenorrhoea. They were all male and had 4 lower limbs, 4 upper limbs, and two distinct well-positioned anuses [Figure 2]. The umbilical cord was single [Figure 2 and Figure 3]. The birth weight was 7 thousand grams. No evidence of meconium emission was observed. The presence of a serosanguineous bump on inspection and palpation was noted. The ears were large. The neonatal evolution was poor because both twins died five minutes after delivery in a respiratory distress situation. The absence of oxygen in the medical environment and resuscitation equipment did not allow for good resuscitation of the twins. The informed consent of the family was not obtained to perform the dissection and anatomical observations.



Figure 1. View of conjoined twins after extraction during cesarean section.



Figure 2. High view of conjoined twins, showing the fusion of two abdomens and thoraxes.



Figure 3. View of the single umbilical cord, between the two conjoined twins.

It should be noted that after the extraction of the twins, the transfusion of 450 millilitres of blood was set up. APGAR's score in the first minute was three points.

3. Discussion

Human embryogenesis, which covers the period from fertilization to segmenta-

tion, blastulation, implantation, and gastrulation, is essential for the compression of congenital malformations.

In the case of twin pregnancy, under normal conditions, the embryo undergoes successive and complete divisions depending on the fertilized ovules: two fertilized ovules result in dizygotic twin pregnancy, while the fertilization of a single ovule requires segmentation and is monozygotic. Depending on the period of segmentation, it is either a dichorionic-diamniotic pregnancy (beginning of segmentation), or a monochorionic-diamniotic pregnancy (embryonic button stage), or a mono-chorionic-mono-amniotic pregnancy (embryonic disc stage). Twins pregnancy is therefore characterized by two embryos that evolve simultaneously and its frequency is increasing, as in all cases of multiple pregnancies, following the decrease in the age of primiparity and the use of modern techniques of medical assistance in procreation (AMP) [13] [14]. This constitutes a risk of morbidity and mortality of newborns due to respiratory distress, prematurity, congenital malformations, etc. [14].

The first twins, in the mystical and religious conscience, have divine and mystical powers. This belief has led the followers to various beliefs, which are transmitted from generation to generation, with the possibility of sacrifices and specific ceremonies [14] [15]. In some villages in Lubao territory (such as Kaombo), twin villages are held.

The conjoined twins are witnesses to an error of incomplete division of the monozygotic embryonic disc cells (as a rule) with the consequence of congenital dysmorphism by sharing of organs [2] [3] [10] [16]. But the unanimity is not yet established of the fact that the mechanism is poorly elucidated [7]. The majority of the authors evoke the secondary fusion of the already separated discs or the embryonic fission which results in the absence of separation of the twins during the embryonic period, but their origin is not yet elucidated [4] [17] [18]. However, it has been pointed out that the phenomenon occurs after the thirteenth-day post-conception, essentially between the 15th and 17th day [2] [3].

The first mention of “Siamese” twins is recorded in Neolithic mythology, where the mystic-religious problem of “monster twins” was addressed [18]. It is in Armenia that the first description of conjoined twins dates back to 945 by Léon Le Diacre: the twins of Constantinople, in Asia Minor [19]. These twins were thoracic and male and died three days after their separation by the Byzantine surgeons [19]. It was not until the 17th century, in 1689, that the first success was achieved with the surgical separation of conjoined twins by the Swiss Johannes Fatio.

The name “Siamese”, commonly used to designate conjoined twins, comes from the Kingdom of Siam (now Thailand), where the most famous conjoined twins in history were born in 1811: the Eng and Chang Bunker brothers, businessmen and fathers of twenty-one children [2].

From the tenth to the beginning of the twentieth century, conjoined twins were wrongly called “monsters”. They were presented to the public, sometimes

to obtain money: the case of the “Orissa sisters” Radica and Doodica, and the sisters Rosa and Joseph Blazheks in Europe and the United States. Moreover, this embryological anomaly was associated with the curse, such as the aforementioned Rosa and Joseph Blazheks sisters, born in 1878 to a young woman of 22 years of age who “attracted the curse by looking at cripples with bad intentions” according to detractors, which contributed to forbidding pregnant women from looking at these handicapped people [20].

In the scientific literature, the incidence of conjoined twins, all forms combined, is from one case in ten thousand to 250 thousand [1] [2] [4] [5] [6] [7].

It has been shown that the majority of conjoined twins, 90% - 95%, are female, although the male sex predominates among the cases of monozygotic pregnancies [2] [4]. In the latter case, there is no scientific explanation put forward to date, but we believe that the genetic theory is plausible. The conjoined twins in this study were all male. In addition, the presence of a single placenta and a single umbilical cord (monomphalic twins), suggest monozygotic, as do the sisters and brothers Abigail and Brittany Hensel, Eng and Chang Bunker, Rosa and Joseph Blazheks.

However, as we have pointed out in the pathogenic mechanism (fusion and fission), it is not absolute that all conjoined twins are of the same sex: there are exceptional cases of opposite sexes, for example, the conjoined twins George and Lori Schappel, and that in addition, in triplet pregnancy, ante-natal selection can be made according to the possibility of survival [6].

The diagnosis can be in the antepartum period or during delivery, as in our context. Ultrasound and magnetic resonance imaging (MRI) are used. Abdominal or transvaginal ultrasound is effective in making the diagnosis as early as the seventh week of amenorrhea [21] [22]. The advantages of prenatal diagnosis are enormous, as it can facilitate the selection of embryos, as mentioned by Liu *et al.* [6] by injection of potassium chloride in Intra thoracic.

The disadvantage of diagnosis at the time of delivery is no longer to be demonstrated because of the risks associated with Caesarean section. In our case, the C-section incision had to be enlarged to allow extraction of the twins. In addition, it was a surprise for the surgical and anaesthetic teams.

The classification of conjoined twins based on the arrangement of body parts includes symmetric and asymmetric twins. Symmetrical twins can be in a parapagus, dorsal, or mostly ventral position. The ventral symmetrical position includes twins joined by the abdominal region (omphalopagus), thoracic (thoracopagus), the fusion of two heads into one (Cephalopages), or sometimes ventral and caudal fusion through the hips (Ischiopages), etc. There are mixed forms, cephalothoracopagus or thoraco-omphalopagus called tronchopagus [3] [6] [8] [12].

Our series of study, it is about the joint twins thoraco-omphalopagus or Tronchopages [5] [16], that is to say, that the chest and the abdomen were joined in both. It is a less serious form, especially if the vital organs are not common [3].

Conjoined twins, in addition to the complications related to twinhood, have a

poor prognosis [5] [6] [14].

However, the prognosis depends on the degree of fusion, the severity of organ involvement, and especially in case of associated congenital abnormalities: liver, heart, kidney, etc. [3] [8].

Hence, the majority of deaths were observed in utero or during the neonatal period [3] [16] [17] [23]. In our study, the twins died too early, five minutes after birth, far from the one hour mentioned by Kattel *et al.* [23] in the context of poor APGAR score (three points) and lack of resuscitation means, apart from oxygen supplementation. In the case of our conjoined twins, we believe that the combination of several factors, including maternal anaemia, acute fetal distress, and lack of adequate resuscitation equipment, is responsible. In the history of the Democratic Republic of the Congo, there is no such case of separation of conjoined twins. The means must be implemented to improve the share of neurosurgery in provincial hospitals, Lubao being located more than 1300 kilometers from the capital Kinshasa where one can meet the few neurosurgeons.

Table 1. Information on African studies of conjoined twins.

No	Authors	Country	Year	Types of conjoined twins	Time of diagnosis	Comment
1	Chrysostome <i>et al.</i> [25]	CAR*	2021	Craniopagus	At the birth	Joined twins successfully separated in Rome (Italy) after 30 hours of surgery
2	Konan <i>et al.</i> [26]	Ivory Coast	2008	Craniopagus	At the 37th week of amenorrhea	No separation performed due to lack of appropriate means to perform the surgery
3	Zanga <i>et al.</i> [27]	Burkina Faso	2018	Thoracopagus	At the birth	Joined twins died of infanticide on the day of surgery
4	Osman <i>et al.</i> [28]	Morocco	2012	Omphalo-Ischiopagus	At the 14th week of amenorrhea, by ultrasound	Termination of pregnancy after parental consent
5	Iman <i>et al.</i> [29]	Morocco	2021	Parapagus dicephalic	In the 2nd trimester of pregnancy	Conjoined twins born by cesarean section: two heads, three upper limbs and two lower limbs
6	Kheir <i>et al.</i> [10]	Sudan	2012	Craniopagus	In the 1st trimester of pregnancy	Successful separation
7	Swar <i>et al.</i> [30]	Sudan	2011	Dicephalus tribrachius	At delivery by cesarean section	Deaths born, probably as a result of poor conditions during pregnancy and the perinatal period
8	Our study	DRC (Kalemie)	2022	Thoraco-omphalopagus	At delivery by cesarean section	Conjoined twins who did not survive more than five minutes after delivery

*CAR: Central African Republic.

Research must be undertaken to provide explanations for this aberrant embryogenesis and at best to avoid them or to facilitate the survival of conjoined twins. It is not surprising that conjoined twins live a long time, without being separated, and contribute to a harmonious life in society. There are many examples in the general literature: Mary and Elyza Chulkhurst (1100-1134), Rosa and Joseph Blazheks (1878-1922), Lori and George Chappell (1961 to present), Abigail and Brittany Hensel (1990 to present, schoolteachers), Chang and Eng Bunker (1811-1874), Ladan and Laleh Bijani (1974-2003), Daisy and Violet Hilton of Brighton (1908-1969), Yvonne and Yvette Mc Carther of Los Angeles, etc. [24]. **Table 1** shows the information on African studies of conjoined twins.

4. Conclusion

Monozygotic and monochorionic twin pregnancy is an anomaly of embryonic development that can result in conjoined twins. The origin of conjoined twins is not mystical or religious. Two mechanisms have been suggested in the literature: fusion and fission of the embryonic disc, between the 15th and 17th day after fertilization. The prevalence of conjoined twins is still unknown in the Democratic Republic of the Congo. According to the scientific literature, this study appears to be the first description. In the context of our study, the conjoined twins were monomphalic and thoraco-omphalopagus, premature, and born by cesarean section. The evolutionary picture was characterized by an early death in a picture of respiratory distress. To this end, the requirement in favour of equipping reference hospitals with qualified human resources (resuscitation anaesthetists and surgeons) and resuscitation and diagnostic materials remains paramount. Ultrasound remains the main means of in utero diagnosis, and pregnant women should be encouraged to undergo at least three ultrasound examinations during the gestational period.

Authors' Contributions

All the authors of this article contributed to the conception, writing, and validation of the manuscript. They have read and approved the final version submitted for publication.

Conflicts of Interest

The authors declare no conflict of interest concerning this study.

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