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Difficulties in Diagnosis and Management of Conjoined Twins: 5 Cases Report in an African Environment (Côte d'Ivoire)/Challenges in Diagnosis and Management of Conjoined Twins: A Five Cases Series in an Africa Setting (Ivory Coast)

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Abstract

Context: The presence of conjoined twins is a rare occurrence during pregnancy. Today, early diagnosis is possible thanks to advances in imaging, particularly three-dimensional ultrasound, and the intervention of a trained operator. This antenatal diagnosis can be used to decide whether or not to continue the pregnancy. **Cases presentation:** Based on the authors' experience, five (05) cases of conjoined twins were collected over a period of six years (01 January 2018-01 January 2023) in the maternity wards of the Angre's Teaching Hospital (study location), whose diagnosis was sometimes unexpected in the delivery room. Among the five cases, we described three (03) thoraco-omphalopages and two (02) parapages, with a female predominance of 4/5. None were candidates for separation because they were stillborn or died on the first day of life. **Conclusion:** This series of cases highlights the different characteristics of conjoined twins and the poor fetal prognosis due to diagnostic difficulties and therapeutic possibilities that are practically non-existent in the African environment.

Keywords

Conjoined Twins, Intra Uterine Fetal Death, Antenatal Diagnosis

1. Introduction

Conjoined twins are one of the rarest congenital anomalies. Their frequency is estimated at 1/50,000 to 1/200,000 births [1] [2]. Untimely vaginal delivery of conjoined twins is a rare event that can be life-threatening for the mother [3]. Today, ultrasound can diagnose this exceptional phenomenon from the first trimester of pregnancy and specify the site of union and frequently associated malformations [4]. The fact that many women do not undergo ultrasound during gestation in our environment, due to the inaccessibility of the technical facilities, exposes practitioners to such an eventuality [5]. We report here 5 cases of conjoined twins whose diagnosis was sometimes made incidentally after delivery.

2. Clinical Cases

These were 5 young patients seen at the Teaching Hospital of Angre in Abidjan, aged between 21 and 24, with no evidence of consanguinity or use of teratogenic substances. They had no occupation or were housewives with a low socioeconomic level. Antenatal follow-up was not optimal during the pregnancy; they had 2 to 4 antenatal consultations, carried out by a midwife, with very few blood tests.

In the first observation, the 21-year-old pregnant woman, G2P0, had undergone 2 ultrasound scans, the first at 29 weeks' amenorrhoea, which revealed a twin bichorionic biamniotic pregnancy with no detectable anomalies, and the second at 32 weeks' amenorrhoea, which revealed a twin monochorionic monoamniotic pregnancy with no anomalies in the foetuses. The 2 ultrasounds did not reveal any malformations. On admission to the delivery room, it was concluded that the active phase of labour had begun in a twin pregnancy of 32 weeks + 4 days with intra uterine fetal death. The evolution was marked by the expulsion of twin parapagal dicephalic macerated stillborn female twins, with a single weight of 2500 g, height = 40 cm, head circumference at 30 and 31 cm, after episiotomy. These were conjoined twins with a single trunk and 2 different heads (**Figure 1**); the umbilical cord was single (dicephale parapagus twins).

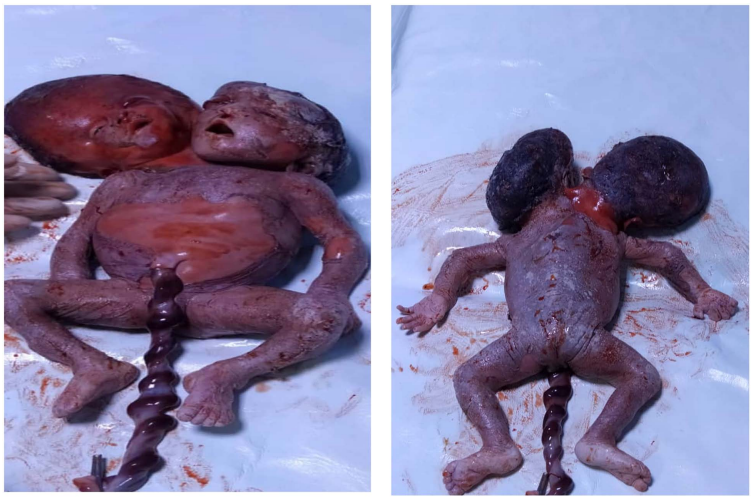

Figure 1. Appearance of macerated stillborn parapagus twins: front and back views.

The 2nd observation concerned a 21-year-old primigravida woman who had consulted a doctor because of dyspnoea.

Ultrasound showed hydramnios (AHI = 32 cm) and an evolving 26-week monochorionic monoamniotic twin pregnancy with two symmetrically attached fetuses in mirror image (Figure 2). The 3D reconstruction showed the areas of attachment on the thorax and abdomen (Figure 3). The twins shared the same heart and liver. A caesarean section was performed to extract two dead female foetuses weighing a total of 2650 grams, joined at the thorax and abdomen by a single umbilical cord containing six blood vessels (Figure 4).

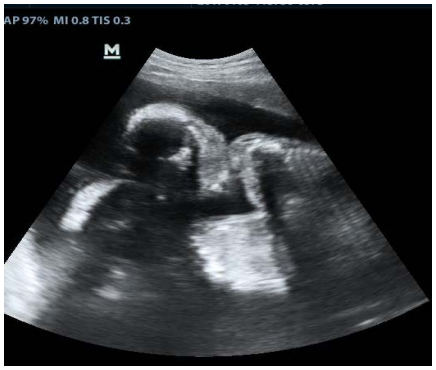

Figure 2. Ultrasonographic appearance of two symmetrical fetuses, in mirror image.

Figure 3. 3D obstetrical ultrasound showing the areas where the thorax and abdomen are joined.

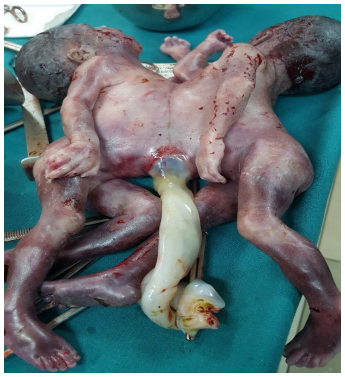

Figure 4. Appearance of stillborn twins at birth.

The 3rd observation concerned a 23-year-old G2P0 patient. She had no medical or surgical history. She was referred from a community health center for severe pre-eclampsia at 34 weeks and 2 days. Early ultrasound at 13 weeks and 3 days showed a triple bichorionic biamniotic pregnancy with conjoined twins in one amniotic sac and a normal fetus in the second sac (Figure 5). Following this consultation, the patient was lost to follow-up and did not attend any further consultations until the 34th week when she presented severe pre-eclampsia in a triple pregnancy. An emergency caesarean section made it possible to extract three foetuses: 2 conjoined male twins, connected by the thorax and abdomen (thoraco-omphalopagus), stillborn and macerated, weighing 1800 g together, and the third live male neonate, weighing 2200 g, height 48 cm, head circumference 31 cm, APGAR 7-8 in 5 minutes (Figure 6). Examination of the live neonate was normal.

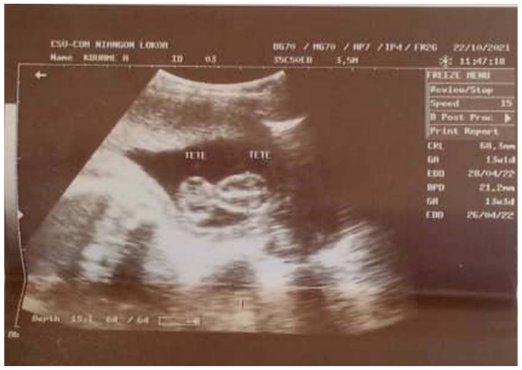

Figure 5. Two conjoined twins in the same pouch (foetal heads at the same level and in the same body plane).

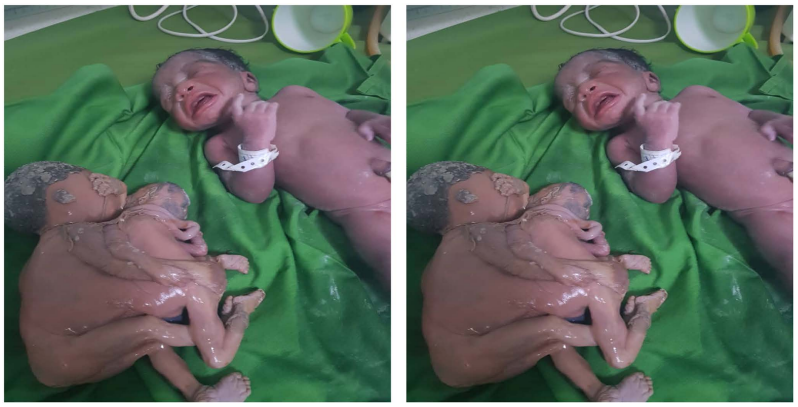

Figure 6. Stillborn thoraco-omphalopagus conjoined twins with macerated weight disproportion next to the apparently healthy 3rd twin.

The 4th observation concerned a 24-year-old G3P1 patient. She had no medical or surgical history. She was referred for perinatal asphyxia in the context of a twin pregnancy at 36 weeks and 3 days. An emergency caesarean section was used to extract 2 conjoined twins with 2 d. The 4th observation concerned a 24-year-old G3P1 patient. She had no medical or surgical history. She was referred for perinatal

asphyxia inistinct heads resting on a single trunk with a single pelvis and 5 limbs (double-headed parapagus), stillborn together weighing 4100 g (**Figure 7**).

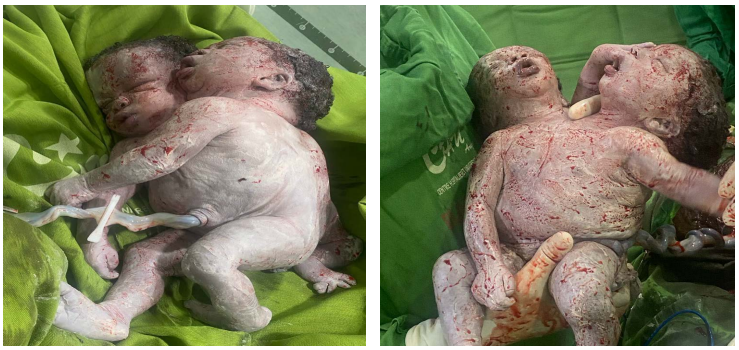

Figure 7. Twins with two-headed parapagus after caesarean section (dicephale parapagus).

The 5th observation concerned a 23-year-old G2P1 patient. She had no previous medical or surgical pathological history. She was referred for stationary dilatation in a twin pregnancy at 34 weeks. No ultrasound scan had been performed. The emergency caesarean section allowed extraction of 2 conjoined female twins joined by the thorax and abdomen, i.e. thoraco-omphalopagus, alive and together weighing 3800 g (**Figure 8**). The twins died after 1 day.

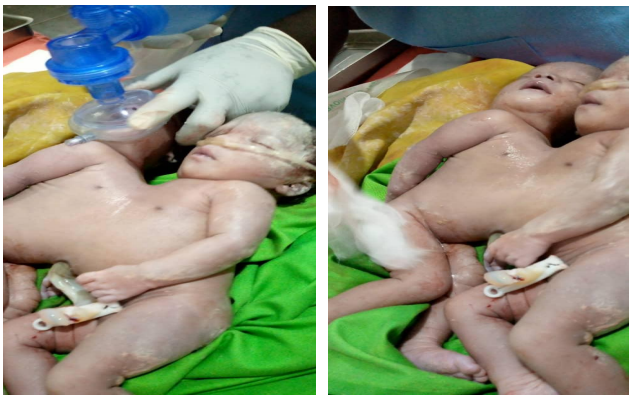

Figure 8. Thoraco-omphalopagus twins.

3. Discussion

Mono-amniotic pregnancies are very rare. They account for 1% of twin pregnancies and are fraught with specific complications, including incomplete division, resulting in conjoined or Siamese twins that are problematic to manage, even for highly qualified teams [6] [7]. The etiopathogenesis of conjoined twins is poorly understood. There is no associated chromosomal abnormality. Race, heredity, parity and consanguinity are not thought to be involved in the process [7]. One of the earliest classifications dates back to 1573 and is attributed to Ambroise Paré [4]. In 1832, Saint-Hilaire established a classification of conjoined twins according to the site of external union and symmetry [3]. Thoracopagus is the most common form, joined by the anterior part of the thorax. The organs are generally asymmetrical.

The pericardium is common for the twins in 90% of cases and the hearts are joined in 75% of cases [3]. These forms are always lethal because scission is impossible. Identification of the common heart, as found in our second observation, darkens the prognosis and early therapeutic termination of pregnancy is also legitimate. Omphalopagus usually has fusion between the xyphoid and the umbilicus [3] [8] [9]. In 50 to 70% of cases, these conjoined twins are thoraco-omphalopagous [10]. The liver is common for the twins in two thirds of cases [3] [11]. In the third observation, the case presented with pre-eclampsia as the risk of pre-eclampsia increases in multiple pregnancies with the number of embryos, according to the literature. The rate is approximately 9% in twin pregnancies, which is 3 times higher than in single pregnancies [12]. Parapagus dicephalic conjoined twins, as in the first and 4th observations, represent 11% of conjoined twins and may have 2, 3 or 4 upper limbs. They are stillborn in most cases because of cardiac and pulmonary malformations [11].

In the age of modern ultrasound, no diagnosis of conjoined twins should come as a surprise at delivery, even in developing countries. Diagnosis of conjoined twins is possible by ultrasound before the 12th week of amenorrhoea. And when there is no possibility of separating them, the pregnancy is generally terminated to prevent complications from a laborious vaginal delivery [6] [13] [14]. An ultrasound scan is often prescribed but not always performed by qualified personnel. This is a frequent situation in our underdeveloped African context. A recent survey of obstetric ultrasound knowledge, attitudes and practices in Senegal revealed that 66.6% of practitioners (doctors and midwives) did not have a diploma in ultrasound [7].

In the literature, the visualisation of two stomachs or two hearts within the same mass confirms the diagnosis. Other signs suggest the existence of conjoined twins, such as the detection of more than three vessels within the single umbilical cord (observation 2), the bifid appearance of the fetal pole in the first trimester, and fetal heads that remain at the same level, the failure of one fetus to change position relative to the other over time [8] [15], the presence of hydramnios which is seen in 50% to 76% of conjoined twins, and the face-to-face presentation of the twins [15]-[17]. A detailed ultrasound scan at 20 days' gestation will define the extent of the conjoined zone and the shared organs. Therefore, the diagnosis should be made with caution in the first trimester, and follow-up imaging should be performed to confirm the diagnosis [9].

The vaginal delivery of a double term monster is therefore seen when the woman has not had an ultrasound scan, generally in underdeveloped countries such as sub-Saharan Africa. Apart from cases of prematurity, death in utero or low birth weight, where delivery can be vaginal, a caesarean section is generally indicated [18]. In the case of the patient in our first observation, although she had given birth vaginally, we did not observe any particular difficulties or serious soft tissue lesions, as the fetuses were stillborn and premature.

In our case study, from 2018 to 2023, we collected 3 thoraco-omphalopages, 2

parapages, with a female predominance of 4/5. None were candidates for separation because they were stillborn or died on the first day of life. Based on data in the literature, the incidence of conjoined twins is between 1/50,000 and 200,000 live births. The thoracopagus form is the most frequent variant, with a predominance of females [8]. In the Brazilian series of 44 cases, 72.5% were thoracophagous, 12.5% paraphagous, 7.5% omphalo-ischiophagous, 5.0% omphalophagous and 2.5% cephalophagous [18].

Preoperative investigations and good planning by a multidisciplinary team are necessary for successful surgical separation. A team with long and extensive experience and a well-equipped centré are essential in order to meet the challenge presented by this anomaly [6]. These conditions are hardly met in the African environment, particularly in sub-Saharan Africa. In industrialised countries, the number of conjoined twins being managed has increased due to the possibilitý of ensuring a normal life for these twins [6] [18].

Three-dimensional ultrasound, Doppler and the application of MRI allow better exploration and make a considerable contribution to antenatal diagnosis [19] [20]. Fetal echocardiography is also of vital importance for Siamese twins with cardiac fusion. Increased levels of alphafetoprotein were reported to be indicative of Siamese twins [21], but data on the specificity and sensitivity of this biochemical marker are lacking. Antenatal diagnosis is useful firstly to inform the parents and also to plan the time, place and method of delivery, as well as planning any possible postnatal separation surgery. In general, it is estimated that the majority of Siamese twins are premature, 40% are stillborn and 35% die within 24 hours of delivery [22]. At present, antenatal diagnosis of Siamese foetuses poses an ethical problem: can this pregnancy have a favourable long-term outcome, leading to the birth of human beings who, after complex separation surgery, will have acceptable development and quality of life, or must such a pregnancy be terminated before reaching the stage of viability?

Treatment requires multidisciplinary expertise and additional costs that very few African households can bear, often with disappointing results. Cases of twins who do not share vital organs such as the heart or brain, such as omphalopagus and pygopagus, have higher survival rates. However, it is difficult to carry out a morphological study to assess associated malformations in the African environment.

4. Conclusion

The management of conjoined twins can only be improved if antenatal diagnosis is carried out to identify common anatomical structures, search for associated congenital anomalies, schedule delivery in a suitable facility and plan multidisciplinary neonatal care. In Africa, all women should have at least one morphological obstetric ultrasound performed by qualified personnel. The lack of state-of-the-art technical facilities and the high cost of postnatal care limit the therapeutic possibilities in Africa.

Authors' Contributions

All the authors contributed to the drafting of this manuscript and read and approved the final version.

Conflicts of Interest

The authors declare no conflict of interest.

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