



Case report

Intraoperative finding of papyraceous twin fetus in pregnant woman without specialized follow-up: A summary of difficulties in accessing prenatal services in low- and middle-income countries

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ABSTRACT

Introduction and importance: The papyraceous fetus is the product of the intrauterine fetal death of a twin in early pregnancy, with retention of the fetus for a minimum of 10 weeks, resulting in mechanical compression of the small fetus. It is a finding that can be found early by ultrasound to avoid increased obstetric risk, but in low- to middle-income countries it is not always available.

Case presentation: We present the case of a 22-year-old multipara patient who was remitted to the emergency department due to a one-hour history of premature rupture of the membranes. Only one control ultrasound was performed at 25 weeks of pregnancy. Cesarean section was performed and two products were found, one of them a papyraceous fetus.

Clinical discussion: Almost 50% of women in low- and middle-income countries don't receive adequate antenatal care. It has been estimated that up to half of twins and almost all triplets are born premature and have a higher risk of dying compared to those born at term; specifically, this risk is much higher if the pregnancy is accompanied by rare pathological conditions.

Conclusion: The papyraceous fetus is a rare condition that represents a risk to the health of the mother and other babies, in case of multiple pregnancy; but it can be diagnosed early through imaging studies. Socio-cultural, socio-economic and direct difficulties of the health system may be the cause of the delay in the diagnosis of this pathological entity, making it an unexpected finding during delivery or intraoperatively.

1. Introduction

Mainly twin pregnancies are associated with greater risk for both the mother and the fetus, when compared with singleton pregnancy [1]. Fetal death is defined by the World Health Organization (WHO) by gestation periods, early fetal death is that which occurs before 20 weeks of gestation, the intermediate between 20 and 27 weeks of gestation and the late or also called death is the one that occurs after the 28th week of gestation [2]. Single intrauterine stillbirth (sIUFD) occurs in approximately 6% of twin pregnancies, making it a common adverse event [1]. Many are first trimester fetal losses, but sIUFD after 14 weeks gestation is associated with preterm delivery with long-term risk of postnatal

brain injury [3].

The papyraceous fetus is the product of the intrauterine fetal death of a twin in early pregnancy, with retention of the fetus for a minimum of 10 weeks, resulting in mechanical compression of the small fetus, experiencing dehydration that resembles that of parchment paper, it is considered a rare complication with an incidence of 1:12,000 pregnancies and between 1:184 and 1:200 twin pregnancies [4]; its etiology is unknown, but it is associated with twin-to-twin transfusion (TTTS), genetic abnormalities or fetal chromosomes and improper cord implantation, such as veiled cord insertion [4].

Papillary fetus is a finding that can be found early by ultrasound to avoid increased obstetric risk, but due to the health inequities that

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currently exist in countries with limited resources or socio-cultural barriers, it has become a late finding, even intraoperatively or during delivery. Based on the above, it is important to perform specialized follow-up during prenatal care in order to reduce morbidity and mortality in the event of these conditions [5]. A clinical case of a papiraceous twin fetus in a pregnant woman without specialized follow-up is described. Therefore, the aim of this report is to raise awareness of this fetal anomaly, its risks and the importance of identifying and breaking down barriers to obstetric monitoring in low- and middle-income countries. This case report followed the SCARE guidelines for its realization [6].

2. Presentation of case

A 22-year-old multipara patient (third gestation; delivery 0; abortion 0 and second cesarean section) was remitted to the emergency department due to a one-hour history of premature rupture of the membranes. Last cesarean section 16 months ago. Only one control ultrasound was performed at 25 weeks of pregnancy. On admission with normal vital

signs. Obstetric examination revealed a gravid abdomen with a height of the uterine fundus of 35 cm, single product cephalic presentation with fetal heart rate was auscultated at 150 beats/min and labor in latent phase. In the vaginal examination, a 1 cm permeable cervix was found, amniotic fluid leakage with grade I meconium. Admission laboratories: Hemoglobin 10.9 g/dl, Hematocrit 31.7% leukocytes $10,660 \times \text{mm}$ and platelets $298,000 \times \text{mm}$. The patient is taken to an emergency cesarean section with the diagnoses of 39-week pregnancy by ultrasound with early rupture of membranes of 1 h of evolution, iterative cesarean section, short intergenetic period, anemia, unfavorable socioeconomic condition and satisfied parity. Cesarean section is performed without complications.

In the surgical findings there were two products. The first living twin, cephalic, female, APGAR score 9/1 min and 10/5 min, weight of 2735 g and size 46 cm; the second twin was described as a papyraceous fetus of around 20 weeks, weight 150 g, height 24 cm, and atrophic placenta (Figs. 1-2). The normal placenta, the papyraceous fetus and the atrophic placenta were sent to the pathology department. The pediatrician finds a healthy newborn with adequate weight. Hospital discharge of mother



Fig. 1. Photograph showing placenta with ischemic amniotic membrane and umbilical cord.



Fig. 2. Intraoperative photograph showing papyraceous fetus and atrophic placenta.

and newborn at 24 h. The histopathology result reports bichorionic bi-amniotic twin pregnancy, with a papyraceous fetus of 18 ± 2 weeks of gestational age, of undetermined sex with signs of intrauterine death estimated to be greater than one week with placenta, amniotic membranes and umbilical cord with chronic ischemic changes.

3. Discussion

Almost 50% of women in low- and middle-income countries (LMICs) don't receive adequate antenatal care [7]. It has been estimated that up to half of twins and almost all triplets are born premature and have a higher risk of dying compared to those born at term; specifically, this risk is much higher if the pregnancy is accompanied by rare pathological conditions [2]. In LMICs, the risk of neonatal death in multiple pregnancies is up to six times higher in the neonatal period (3 to 15 days) and up to twice as high in the postneonatal period [2]. However, in general, the neonatal mortality rate in Latin America ranges between 20 and 39 per 1000 live births, and the perinatal mortality rate in the same proportion, very close to that of Africa [2]. Currently there is no study

reporting the incidence of papyraceous fetus in Latin America, therefore, it is not possible to determine in detail the impact of this condition on the burden of obstetric disease in this region.

The specialized follow-up in this type of conditions lies not only in the imaging evaluation, but also in the early diagnosis of disorders that influence fetal growth that may trigger similar disorders in the fetus, such as umbilical cord anomalies, hypertensive disorders, antepartum hemorrhage, premature labor, and anemia [8,9]. However, in the case of the papyraceous fetus, the diagnosis is purely imaging [10]. Many difficulties impede access to prenatal services in LMICs (Table 1) [7,11–19]. In this case, due to the socioeconomic condition, having access only to a first level care center (where there is no control or specialized management of obstetric pathologies), and being in a very distant rural area, were the main causes of the early diagnosis and adequate approach, putting the mother and the other twin at risk.

Although the WHO [2] and scientific societies of gynecology and obstetrics around the world [4] have set as one of the main objectives the care of the pregnant woman and the baby through the prenatal control, it is observable the wide gap between what is expected and what

Table 1

Summary of difficulties in accessing prenatal services in low- and middle-income countries [7,11–19].

Socio-cultural reasons
Absence of signs/symptoms during pregnancy
Keeping pregnancy a secret
Lack of knowledge about the benefits of prenatal care
Embarrassment
Preferences regarding the use of traditional medicine and healers
Fear of the impact of technological tools on the health of the fetus
Resistance and influence by family members
Laziness
Popular beliefs about medical care (harm to the baby)
Mother's low education level
Late initiation of prenatal care
Lack of women's autonomy (domestic violence - dysfunctional household)
Socio-economic reasons
Limited resources for basic consumption in the home
Lack of transportation or high cost of transportation
Long distances to high-level hospitals
Restrictions at work to attend control appointments
Care of other children in the case of multiparous children
Belonging to the rural area
Direct health system reasons
Inadequate hospital infrastructure
Presence of first level hospitals in difficult to access areas
Health caregivers not trained in obstetrical care
Waiting time for service
Negative attitude of the medical team towards pregnant women
Lack of drugs
Lack of medical supplies
Lack of technological equipment and access to the Internet
Insensitivity, disrespect and abuse by health care providers
Inflexibility in scheduling (delay in granting medical appointments).
Low funding for prenatal care campaigns
Corruption

is real. More broadly, the United Nations (UN) stipulated 17 sustainable development goals to be achieved by 2030 [20], within which poverty, health and well-being, reduction of inequalities and sustainable communities and cities [20], were involved in this case, and were the determinants of the absence of access to quality health services that would reduce the risk of obstetric complications arising from the presence of a papyraceous fetus, such as dysostosis, obstruction during labor, aplasia cutis congenita among others [21].

Higashi et al. [22] and Moran et al. [23] studied the impact of obstetric morbidity and mortality in LMICs on the burden of disease and costs to health systems, respectively, showing that obstetric conditions such as maternal hemorrhage, obstructed labor, obstetric fistula and abortion, which are surgically resolved, generate 56.6 million disability-adjusted life years (DALYs) [22]. Neonatal encephalopathy (16.2 million DALYs) and abortion (2.1 million DALYs) are two of the conditions that generate the greatest burden, and these are preventable in a large number of cases [22]. Regarding costs, gestational diabetes, obesity, neuropsychiatric disorders, hypertensive disorders, epilepsy and domestic violence can generate up to \$10,000 per case, and these are also preventable and treatable conditions in early pregnancy [23].

Unlike the cases reported for papyrus fetus, this is the only one that speaks not only of the infrequency of this entity, but also of the difficulties in terms of access to obstetric health services, social inequities and the economic-morbidity-mortality impact. Therefore, it is relevant to suggest strategies for the improvement of programs for pregnant women, such as well-developed prenatal check-ups and access not only to patients in urban areas but also in rural areas, so that there is more access and there are no cases of pathologies that are only diagnosed or

treated at the time of delivery or intraoperatively. Health systems are responsible for guaranteeing access to health services, as well as promoting health education to reduce disease burden indicators and improve the quality of life of the population. This case also demonstrates the current barriers and challenges that need to be overcome in low- and middle-income countries. The patient was satisfied with the approach and regretted the difficulties of not having timely access to prenatal screening.

4. Conclusion

The papyraceous fetus is a rare condition that represents a risk to the health of the mother and other babies, in case of multiple pregnancy; but it can be diagnosed early through imaging studies. Socio-cultural, socio-economic and direct difficulties of the health system may be the cause of the delay in the diagnosis of this pathological entity, making it an unexpected finding during delivery or intraoperatively.

Abbreviations

DALYs	disability-adjusted life years
LMICs	low- and middle-income countries
SIUFD	single intrauterine stillbirth
TTS	twin-to-twin transfusion
UN	United Nations
WHO	World Health Organization

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Ethical approval

Hospital exempts ethics approval for reported cases.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors equally contributed to the analysis and writing of the manuscript.

Research registration

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Declaration of competing interest

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