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FETAL DEATH DUE TO GASTROSCHISIS ASSOCIATED WITH INTRAUTERINE GROWTH RESTRICTION. CASE REPORT

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Palabras clave: Gastrosquisis; Retardo del Crecimiento Fetal; Muerte Fetal.

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RESUMEN

Introducción. La gastrosquisis es un defecto congénito de la pared abdominal que tiene una baja prevalencia y cuya etiología aún es poco conocida. En la mayoría de los casos, el pronóstico es favorable si se detecta de forma temprana y la mortalidad asociada suele ser baja si se hace un seguimiento adecuado; sin embargo, cuando el defecto está asociado a restricción del crecimiento intrauterino (RCIU) el riesgo de desenlace perinatal adverso aumenta.
Presentación del caso. Joven de 16 años con embarazo de 34.6 semanas, quien ingresó en fase expulsiva del trabajo de parto al servicio de urgencias del Hospital Simón Bolívar E.S.E. de Bogotá (Colombia). La paciente reportó hallazgo ecográfico de gastrosquisis y RCIU, condiciones que causaron muerte fetal.
Conclusiones. La identificación y atención de la gastrosquisis es necesaria, no solo para garantizar un desenlace compatible con la vida del feto, sino para lograr el bienestar y las condiciones dignas para el recién nacido ya que la complejidad de esta condición dificulta la obtención de resultados perinatales satisfactorios. El presente caso resalta la importancia de, por un lado, iniciar de forma temprana un control prenatal adecuado para identificar alteraciones de la gestación y, por el otro, prevenir el embarazo adolescente dadas las complicaciones y los desenlaces adversos que se pueden presentar en este.

ABSTRACT

Introduction: Gastroschisis is a congenital defect of the abdominal wall that has a low prevalence and a poorly understood etiology. In most cases, its prognosis is favorable if detected early, and the associated mortality is usually low if adequate follow-up is performed. However, when the defect is associated with intrauterine growth restriction (IUGR), the risk of adverse perinatal outcome increases.
Case presentation: A 16-year-old female, 34.6 weeks pregnant, was admitted to the emergency department of the Hospital Simón Bolívar E.S.E. in Bogotá (Colombia) during the third stage of labor. The patient reported ultrasound findings of gastroschisis and IUGR, conditions that caused fetal death.
Conclusions: Identifying and treating gastroschisis is necessary, not only to guarantee an outcome compatible with the life of the fetus, but also to achieve well-being and dignified conditions for the newborn, since the complexity of this condition makes it difficult to obtain satisfactory perinatal outcomes. The present case highlights the importance of early initiation of adequate prenatal care to identify gestational alterations and of preventing teenage pregnancy, given the complications and adverse outcomes associated with this type of pregnancy.

INTRODUCTION

Gastroschisis is a congenital defect characterized by herniation of the abdominal organs into the amniotic cavity through an opening in the abdominal wall. This is one of the most common defects of the abdominal wall, with a prevalence of 1 case per 4 000 live newborns (1). Gastroschisis is more common when mothers are under 20 years of age and usually has a good prognosis, with a survival rate of 90-95% (2). However, morbidity may increase to a range of 38-77% and mortality may be as high as 50% when it is associated with intrauterine growth restriction (IUGR) (3). It is rarely associated with conditions such as aneuploidy and coexisting intestinal anomalies such as intestinal atresia (15%) (2).

Currently, several approaches are available for the follow-up and medical-surgical treatment of neonates with gastroschisis, including primary reduction, staged reduction with closure of the wall without sutures, or sutured closure together with parenteral nutrition (2). The following is a case of fetal death due to gastroschisis associated with IUGR.

CASE PRESENTATION

A 16-year-old primigravid female was admitted to the emergency department of the Hospital Simón Bolívar E.S.E. in Bogotá (Colombia), a tertiary care center, on September 13, 2022, during the third stage of labor. The patient was 34.6 weeks pregnant and in the previous 12 hours had presented with pelvic pain, abundant vaginal discharge, and absence of fetal movements for approximately 3 hours. On admission. She reported a diagnosis of fetal gastroschisis determined on screening ultrasound performed at 14.1 weeks (Figure 1) and stated that a fetal blood flow assessment was performed at 22 weeks, the results of which were within normal values.

Figure 1. Screening ultrasound performed at 14.1 weeks. Intestinal loops are seen floating in the amniotic fluid.

Source: Image obtained while conducting the study.

The patient was underweight throughout her pregnancy, with a body mass index on admission of 18.3kg/m^2. She had no history of alcohol, tobacco or psychoactive substance use, and stated that her menstrual cycles prior to pregnancy were irregular, that she had never undergone a Pap smear or mammogram, and that she had never used contraceptive methods.

During the physical examination on admission, the following findings were reported: abdomen slightly painful on palpation in the lower portion, fundal height of 22cm, normal maternal external genitalia, and a single fetus lying in longitudinal position with cephalic presentation and dorsum to the right. Vaginal examination showed complete dilatation of the cervix (10cm), 100% cervical effacement, fetal head 3cm below the pubic symphysis, and unruptured amniotic membrane.

The results of the first, second and third trimester laboratory tests presented by the patient were within normal ranges. The TORCH screen (toxoplasmosis, rubella cytomegalovirus, herpes simplex, and HIV) performed at 28.6 weeks was negative, and the ultrasound follow-up performed by the maternal-fetal service showed an estimated fetal weight of 967g and percentile <3.

Given the gastroschisis diagnosis, laboratory tests (blood count, hepatitis B surface antigen, treponemal serological test for syphilis, and rapid HIV test [types 1 and 2]) were requested on admission, with normal results. A transabdominal ultrasound was performed to evaluate fetal blood flow, which did not detect fetal heartbeat.

Considering the characteristics of the case, immediately after the ultrasound finding, the patient was transferred to the delivery room and intravenous fluids were started (Ringer's lactate at a dose of 70mL/hour). After 30 minutes of admission, a female stillborn baby weighing 1 920g and measuring 42cm was born; the APGAR test score performed at 09+00 hours on September 13, 2022, was 0-0-0. The physical examination of the stillborn showed generalized skin necrosis, absence of palpable pulse in the umbilical cord, purplish umbilical cord, and abdominal wall defect in the right paraumbilical area with complete prolapse of intestinal loops with a necrotic appearance (Figure 2).

Subsequently, the mother was transferred to the recovery room for puerperium management. There were no maternal complications; however, due to the high thromboembolic risk associated with fetal death, immediate treatment was indicated with subcutaneous enoxaparin 40mg/day, antibiotic prophylaxis with intravenous ampicillin 2g in a single dose, Ringer's lactate at a dose of 70mL/hour in continuous infusion, and hyoscine 20mg every 8 hours for analgesia. After being monitored for 24 hours, the patient was discharged with indication of thromboprophylaxis with 40mg of subcutaneous enoxaparin for 6 weeks, a single dose of Cabergoline 1mg orally for lactation suppression, and analgesic therapy with hyoscine 10mg every 8 hours and acetaminophen 500mg every 6 hours orally for 5 days. Likewise, she was advised to attend a postpartum follow-up appointment after 7 days.

Figure 2. 34.6 week stillborn with evidence of gastroschisis.
Source: Image obtained while conducting the study.

Following the delivery, the fetus was taken to the hospital's pathology department, which then transferred it to forensic medicine, so no autopsy report is available. Regarding the postpartum care of the patient, since the patient decided to make an appointment with her healthcare provider at a different center, it was not possible to follow up on the patient's case.

DISCUSSION

Gastroschisis is a defect that affects the thickness of the abdominal wall around the umbilicus, usually to the right of the insertion of the umbilical cord, which in most cases is <4cm. Herniation of the abdominal organs, mainly the intestine, protrude through this defect into the amniotic cavity without involving the umbilical cord (1).

The embryological origin of this defect is still under discussion. However, among the possible causes are failure of differentiation of the mesenchyme following teratogen exposure, rupture of the amniotic membrane at the base of the umbilical cord, interruption of the omphalomesenteric artery causing localized necrosis of the abdominal wall at the base of the cord, abnormal involution of the right umbilical vein, and abnormal folding of the embryo leading to a ventral body wall defect (4).

Some studies describe various environmental determinants as risk factors for developing gastroschisis. For example, Rodríguez-Acosta *et al.* (5), in a study of 22 women diagnosed with abdominal wall defects, found that the most frequent

risk factors for these defects were a combination of environmental factors, failure to take periconceptional folic acid supplementation, and exposure to chemicals. This same study established a direct relationship between gastroschisis and juvenile maternal age and low weight (5), both of which are characteristics observed in the patient in this report.

Gastroschisis can be simple or complex. In the latter, additional intestinal complications such as atresia, perforation, or necrosis may occur; therefore, it is associated with an increase in morbidity and a reduction in the survival rate, from >90% to 70-80% (6). In the present case, due to its association with IUGR and its fatal outcome, gastroschisis was considered as complex.

The diagnosis of gastroschisis is established when free intestinal loops are observed floating in the amniotic fluid with the umbilical cord correctly inserted. This defect can be identified on ultrasound between weeks 11 and 14 of pregnancy (1), as occurred in the case presented. Once identified, constant monitoring by ultrasound is recommended to evaluate possible complications and outcomes (6).

Cesarean delivery rates due to gastroschisis are high (60% in the United States). However, this defect alone is not an absolute indication for a cesarean section since vaginal delivery has not been shown to be a risk factor for neonatal complications such as intestinal obstruction, sepsis, or necrotizing enterocolitis (7).

In order to reduce perinatal morbidity and mortality, new techniques have been implemented to attend delivery. One of such techniques is the EXIT (Ex-utero intrapartum treatment) procedure, which involves maintaining fetal circulation until an airway is secured, thus guaranteeing the uteroplacental gas exchange necessary to achieve hemodynamic stability in severe conditions of airway compression in the fetus, facilitating a possible immediate surgical procedure (8).

When gastroschisis is diagnosed, it is essential to establish the route of delivery in a timely manner to be able to perform the procedure correctly, but the cesarean section is preferred because it allows maintaining placental circulation. The EXIT procedure is used to reduce adverse perinatal events that may occur during labor, as it allows for controlled management of the fetal airway while maintaining fetal circulation. Furthermore, it reduces respiratory effort and prevents aerophagia, unlike conventional vaginal and cesarean deliveries, thus maintaining optimal fetal hemodynamics while multiple additional procedures are performed (8).

Postnatal care of gastroschisis aims to prevent evaporative fluid loss, hypothermia, and possible infections. Surgical treatment seeks to achieve timely reduction of the herniated organs and is performed using two major techniques: primary reduction and staged reduction, the latter being more recommended because it reduces the duration of mechanical ventilation, the risk of infection, and the time to enteral nutrition (6).

It has also been established that a small-for-gestational-age fetus is a fetus

whose estimated fetal weight is below the 10th percentile for gestational age but has ultrasound findings of blood flow that are within normal limits. On the other hand, IUGR is defined as a condition in which the estimated fetal weight is below the 10th percentile for gestational age and the ultrasound shows signs of fetal involvement due to abnormalities in the evaluation of fetal and placental blood flow or an abdominal circumference below the 3rd percentile for any gestational age. The main difference between these two conditions is the increased mortality risk, which is higher in fetuses with IUGR (9). The risk of fetal death is approximately 1.5% when the estimated fetal weight is below the 10th percentile for gestational age and increases to 2.5% when it is below the 5th percentile (10).

IUGR is classified into early and late depending on the time of diagnosis: early IUGR, which has a prevalence of 30%, is identified before 32 weeks of gestation, while late IUGR, which has a prevalence of 70%, is identified at 32 weeks or later (9). Perinatal mortality is higher in individuals with early IUGR, which is consistent with the case presented, where it was possible to establish that the estimated fetal weight at 28 weeks of gestation was 967g, with a percentile <3.

IUGR is a multifactorial condition with various etiologies (maternal, fetal, and placental), with placental insufficiency affecting fetal perfusion and nutrition being the most important. In the present case, maternal risk factors included nulliparity, teenage pregnancy, and the low weight of the mother. Moreover, among the genetic and fetal structural factors, gastroschisis was identified as an associated condition because it increases the risk of IUGR by 25% (10).

One of the risk factors associated with IUGR is maternal age. In this regard, Marín-Montoya (11), in a study carried out with the records of 3 428 births assisted between January and December 2011 at the Instituto Materno Infantil Hospital La Victoria in Bogotá, found that 2.47% (n=87) of the newborns were diagnosed with IUGR and that the age of the mother was between 17 and 34 years of age in 86.2% of those cases. However, the author highlights that there were 13 cases in which the mothers were at extreme ages (<16 and >35 years) and that only 2 cases of small-for-gestational-age fetus were recorded.

The highest prevalence of cases with gastroschisis occurs in young mothers (<20 years), and this factor is of great importance. For example, Pérez-Dajaruch *et al.* (3) reported a case in which the only risk factor for the development of this condition was maternal age (18 years). Considering the above, the prevention of teenage pregnancies is of utmost importance to prevent both gastroschisis and IUGR, two conditions that increase the morbidity and mortality of newborns.

According to the World Health Organization, fetal death is defined as death occurring in late pregnancy, but it should be noted that each country establishes its cut-off point for "late" and it may vary depending on legislation; for example, the cut-off point for fetal death in the United States and Mexico is 20 weeks of gestation (12). Among the most relevant risk factors for fetal death are some

found in the present case, such as gastroschisis associated with IUGR, low maternal weight during the entire pregnancy, and maternal age, which favored the adverse perinatal outcome (12).

In the reported case, the patient had no history of alcohol, tobacco or psychoactive substance use. Considering the abdominal wall defect associated with IUGR and the findings of intestinal necrosis in the fetus, it is possible to infer the unpredictability of some pregnancy complications and the lack of a definitive treatment to contain their outcome. It is therefore essential to perform an adequate ultrasound follow-up and encourage adherence to prenatal check-ups, not only to prevent complications, but also to be able to perform early interventions.

CONCLUSIONS

Gastroschisis is a condition with a good prognosis and a high survival rate if it is diagnosed early and if there is adequate prenatal care. However, in countries with difficult access to healthcare there may be unfavorable outcomes. Moreover, when gastroschisis is related to other conditions such as IUGR, fetal mortality can increase from 38% to 77%.

It is necessary to identify and care for gastroschisis not only to guarantee an outcome compatible with the life of the fetus, but also to achieve well-being and dignified conditions for the newborn, since the complexity of this condition makes it difficult to obtain satisfactory perinatal outcomes. This case highlights the importance of initiating early and adequate prenatal care to identify gestational alterations and of preventing teenage pregnancy, given the complications and adverse outcomes that can occur in this type of pregnancy.

ETHICAL CONSIDERATIONS

For the preparation of this case report, informed consent was obtained from the patient and her legal guardian, who authorized the publication of the clinical details.

CONFLICTS OF INTEREST

None stated by the authors.

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