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Clinical case of feto-fetal transfusion syndrome development in dichorionic twin pregnancies with fused placentas

© Marina N. Mochalova¹, Elena V. Falko², Viktor A. Mudrov¹, Anastasiya Yu. Alekseyeva¹¹ Chita State Medical Academy, Chita, Russia;² Regional Clinical Hospital, Chita, Russia

This article analyzes a clinical case of feto-fetal transfusion syndrome development in dichorionic diamniotic twin pregnancies with fused placentas. Feto-fetal transfusion syndrome is a typical complication of the monochorionic type of placentation, but it is quite rare in the dichorionic type of placentation. In this case, the syndrome development became possible due to the close anatomical location of the placentas, which probably led to their fusion and the development of unbalanced interplacental anastomoses. During the observation of the patient in an antenatal clinic, no ultrasound signs of feto-fetal transfusion syndrome were detected. At a 25⁺⁵-week gestation period, the patient complained of cramping pains in the lower abdomen and liquid discharge from the genital tract. The patient was hospitalized at the second stage of labor and gave birth through the natural birth canal. The first fetus had polyhydramnios, the second one having extremely low water. The first was a premature boy in the occiput posterior position weighing 980 g and 32 cm in height in a state of severe asphyxia with an Apgar score of 1, 4 and 6 points. The second was a premature boy in breech position weighing 490 g and 32 cm in height, also in a state of severe asphyxia with an Apgar score of 1, 4 and 6 points. The first child developed severe multiple organ failure, which caused death on the twelfth day. The second newborn developed respiratory distress followed by death on the second day of the neonatal period.

Keywords: feto-fetal transfusion syndrome; dichorionic pregnancy; multiple pregnancy.

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Клинический случай развития фето-фетального трансфузионного синдрома при сращении плацент у дихориальной двойни

© М.Н. Мочалова¹, Е.В. Фалько², В.А. Мудров¹, А.Ю. Алексеева¹¹ Читинская государственная медицинская академия, Чита, Россия;² Краевая клиническая больница, Чита, Россия

В статье представлен клинический случай развития фето-фетального трансфузионного синдрома у дихориальной диамниотической двойни. Фето-фетальный трансфузионный синдром является характерным осложнением для монохориального типа плацентации и при дихориальном типе плацентации встречается достаточно редко. В данном случае его развитие стало возможным ввиду близкого анатомического расположения плацент, что, вероятно, привело к их сращению и развитию несбалансированных межплацентарных анастомозов. Во время наблюдения пациентки в условиях женской консультации ультразвуковых признаков фето-фетального синдрома выявлено не было. На сроке гестации 25⁺5 недель у пациентки появились схваткообразные боли внизу живота и жидкие выделения из половых путей. Пациентка была госпитализирована в стационар во втором периоде родов и родоразрешена через естественные родовые пути. У первого плода отмечалось выраженное многоводие, у второго — абсолютное маловодие. Первым в затылочном предлежании родился недоношенный мальчик массой 980 г, ростом 32 см в состоянии тяжелой асфиксии с оценкой по шкале Апгар 1 и 4 и 6 баллов; вторым в чисто ягодичном предлежании — недоношенный мальчик массой 490 г, ростом 32 см, также в состоянии тяжелой асфиксии с оценкой по шкале Апгар 1 и 4 и 6 баллов. У первого ребенка развилась тяжелая полиорганная недостаточность, которая стала причиной смерти на двенадцатые сутки. У второго новорожденного развилась выраженная дыхательная недостаточность, в результате которой ребенок погиб на вторые сутки неонатального периода.

Ключевые слова: фето-фетальный трансфузионный синдром; дихориальная беременность; многоплодная беременность.

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BACKGROUND

Feto-fetal transfusion syndrome (FFTS) is a severe complication of multiple pregnancies, in which unbalanced placental anastomoses are formed between the circulatory systems of fetuses [1]. This results in a discharge of blood from one system to another and hypovolemia in the donor fetus and hypervolemia in the recipient fetus, as a consequence [2, 3]. The donor fetus develops chronic hypoxia as a result of insufficient perfusion of oxygenated blood, which leads to fetal growth retardation, oligohydramnios, and anemia. An increase in the volume of circulating blood in the recipient fetus causes cardiomegaly, congestive cardiovascular failure, erythremia, polyhydramnios, and nonimmune hydrops fetalis when the donor fetus dies [4].

FFTS occurs in 10% of multiple pregnancies [5], most often with a monochorionic type of placentation. Cases of FFTS development with dichorionic type of placentation are extremely rare [6]. This situation is possible with placental fusion, when anastomoses are formed between the placenta of the donor fetus and placenta of the recipient fetus [7]. Perinatal mortality in this syndrome is 17%–28% [8], and in the absence of adequate treatment, it can reach 80%–100% according to various authors [8–10].

Development of FFTS is expected in the management of patients with a monochorionic type placentation, with obstetricians-gynecologists focusing on its early diagnostics, which is not the case for dichorionic type placentation. The development of this problem is difficult not only to predict, but also to diagnose since it is not well enough discussed in literature and complication rarely occurs in dichorionic twins. Therefore, FFTS is only detected at later stages of development, leading to perinatal losses [6].

Ultrasound study is the main method for FFTS diagnoses, in which special attention is given to assess the size of the maximum and minimum pockets of amniotic fluid and bladder and dynamics of fetal growth. The first sign of this complication is an amniotic fluid imbalance, namely the presence of a maximum pocket of amniotic fluid of 2.0 cm or less on one side of the amniotic membrane and 8.0 cm or greater on the other. The maximum systolic velocity in the middle cerebral artery is measured to diagnose fetal anemia [11–13].

CLINICAL CASE DESCRIPTION

Patient N. is 32 years old, multigravida, multiparous, married, and was registered for pregnancy in the antenatal clinic from the term of 6–7 weeks of gestation. She attended the antenatal clinic regularly (9 visits). Without contraceptive methods, this is now her fourth pregnancy and

second delivery. Patient history revealed an independent childbirth at term, with a boy weighing 2750 g and 50 cm in height; childbirth and postpartum period proceeded without complications. The patient's obstetric history was complicated by pregnancy termination at a gestational age of 10–11 weeks due to anembryonic gestation and medical abortion at a term of 5–6 weeks of gestation, without complications. This is her fourth pregnancy; according to the patient, the first half of the pregnancy was uneventful. Smoking was noted as a bad habit, which the patient gave up at the beginning of pregnancy. Chronic cervicitis of ureaplasmic etiology and chronic endometritis of unspecified etiology were diagnosed among gynecological diseases. Chronic diseases included chronic gastritis, high myopia, and varicose veins of the lower extremities. The total weight gain during pregnancy was approximately 11 kg.

During the follow-up period in the antenatal clinic, 4 ultrasound examinations were performed, and the presence of dichorionic twins was specified at a gestational age of 4–5 weeks and dichorionic twins were confirmed and localization of both chorions was determined (along the anterior wall of the uterus) at 12–13 weeks. Screening ultrasound examination at 20–21 weeks of gestation showed no signs of FFTS (imbalance of amniotic fluid, difference in fetal size).

Cervicometry revealed an internal os dilatation to 5 mm with a cervical length of 30 mm at 23–24 weeks of gestation. Therefore, low-dose progesterone was prescribed to prevent premature birth.

The patient returned to the antenatal clinic with complaints of cramping pains in the lower abdomen and liquid discharge from the genital tract at 25⁺⁵ weeks of gestation. She was examined by an obstetrician-gynecologist, and was diagnosed with the period 1 of very early preterm delivery at a gestational age of 25–26 weeks; dichorionic-diamniotic twins; antenatal discharge of amniotic fluid; and the patient was referred to a hospital.

The patient was admitted to the perinatal center with complaints of cramping pains in the lower abdomen and liquid discharge from the genital tract. Upon admission, moderate strength contractions were recorded for 60 s, with an interval of 1.5 min; a vaginal examination revealed a complete uterine orifice opening, with the head of the first fetus located in the wide part of the pelvic cavity; the patient was diagnosed with the period 2 of very early preterm delivery at 25⁺⁵ weeks of pregnancy; dichorionic-diamniotic twins; breech presentation of the second fetus; chronic placental insufficiency, subcompensated form; and chronic hypoxia of both fetuses.

The patient was hospitalized in the obstetric department, where continuation of conservative childbirth management was decided. The puerpera reported contractions of strains

nature 20 min after admission, she was then examined in the delivery room, with separated fetal membranes, 5 L of light and odorless amniotic fluid were discharged, and the head of the first fetus entered the narrow part of the pelvic cavity. After 5 min from the appearance of strains, in a vertex presentation, a live premature boy weighing 980 g and 32 cm in height was born, with an Apgar score of 1, 4, and 6 points. Contractions of strains nature reappeared 3 min from the birth of the first fetus. In order to clarify the obstetric situation, a vaginal examination was performed, which revealed the buttocks of the second fetus located at the narrow part of the pelvic cavity, the fetal bladder opened during the examination, practically without amniotic fluid. In a purely breech presentation, the second live premature boy weighing 490 g and 32 cm in height was born 5 min after the birth of the first fetus, with an Apgar score of 1, 4, and 6 points; at the birth of the second fetus, posterior amniotic fluid was not noted. Taking into account the pronounced polyhydramnios (>5 L) in the first fetus, absolute oligohydramnios in the second fetus, the form of chronic placental insufficiency was changed to decompensated. During strains, 10 min after the birth of the second fetus, an afterbirth with a size of $40 \times 26 \times 1.5$ cm was discharged independently, consisting of 2 fused placentas with a large number of petrification and anastomoses between placentas. An FFTS diagnosis was confirmed by macro- and microscopic placental examination.

The postpartum period was uneventful.

The condition of children from birth was assessed as severe due to neurological symptoms and respiratory failure in the presence of deep morphological and functional immaturity, which served as an indication for their transfer to mechanical lung ventilation. In the second newborn, respiratory failure (hypoxemia and hypercapnia) increased, and cardiovascular disorders appeared. On day 2 after birth, hemorrhagic syndrome developed. Despite complex therapy, the child's condition remained severe; death occurred on day 2 of life from respiratory failure. In the first newborn, hypoxemia and hypercapnia also worsened, and therefore parameters of mechanical ventilation were increased. During the first days of life of the first newborn, his condition was extremely unstable, and cardiac arrest was recorded twice. Subsequently, the child's condition remained critical but stable. On day 7 of life, a clinical presentation of pneumothorax was registered, which developed due to a decreased lung compliance in the presence of respiratory distress syndrome and intensified parameters of mechanical ventilation, which aggravated respiratory failure and condition severity. The condition of the first newborn progressively aggravated despite the complex therapy, and the phenomena of respiratory failure increased with the

development of hypoxemia, acidosis, arterial hypotension and poor cerebral perfusion, cerebral ischemia, and episodes of seizures. Under the influence of hypoxia, the permeability of the brain membranes increased, and edema developed. Later, the phenomena of intestinal paresis, probably of central origin, oligo- and anuria, an increase in edematous syndrome were noted. Respiratory distress syndrome, which developed in the presence of respiratory system immaturity and history of asphyxia aggravated the severity of the condition of the newborn, whose death occurred on day 12 of life from progressive multiple organ failure. Autopsy revealed that the cause of death of the first newborn was multiple organ failure and that of the second newborn was respiratory failure.

For postmortem examination, 2 fused placentas of the first and second fetuses, $20 \times 14 \times 1.5$ and $20 \times 12 \times 1.5$ cm in size, respectively, were presented. Attention was drawn to a large number of vessels of various sizes located on the fetal surface of one placenta, passing through the amniotic membranes and connecting to the vessels of the second placenta with the formation of pronounced network of anastomoses. The study confirmed that the placentas were dichorionic. Both placentas were immature, with delayed development of fetal angiogenesis and early involutive-dystrophic changes. Chronic subcompensated placental insufficiency with calcareous inlay of villi was confirmed. Moderate compensatory-adaptive reactions, signs of acute placental insufficiency, and villous edema were noted. The umbilical cords were formed anatomically correct (1 vein, 2 arteries), and vessels were emptied without inflammation. Signs of infection were revealed, including diffuse focal extraplacental membranitis, parietal intervillitis, diffuse focal basal deciduitis, intervillitis, villitis, and marginal placentitis (moderate degree of infection along the mixed pathway). Morphological signs of TORCH infection were registered.

DISCUSSION

Patient diagnosis with dichorionic twins was difficult due to an extremely low incidence of FFTS in dichorionic type of placentation; therefore, treatment and delivery were not performed in a timely manner.

Female patients with multiple pregnancies should be referred to as high-risk group and examined in full in due time, regardless of the type of placentation. Medical practitioners, obstetricians-gynecologists, and sonologists, should aim at early diagnosis of FFTS in this group of patients and remember that dichorionic type placentation does not exclude FFTS development.

Conflict of interest. Authors declare no conflict of interest.

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AUTHORS INFO

***Marina N. Mochalova**, MD, PhD, Assistant Professor;
address: 39A Gorky str., Chita, 672090, Russia;
ORCID: <https://orcid.org/0000-0002-5941-0181>;
e-mail: marina.mochalova@gmail.com

ОБ АВТОРАХ

***Марина Николаевна Мочалова**, канд. мед наук, доцент;
адрес: Россия, 672090, Чита, ул. Горького, д. 39А;
ORCID: <https://orcid.org/0000-0002-5941-0181>;
e-mail: marina.mochalova@gmail.com

AUTHORS INFO

Elena V. Falko, MD;
e-mail: p-2000f@yandex.ru

Viktor A. Mudrov, MD, PhD;
ORCID: <https://orcid.org/0000-0002-5961-5400>;
e-mail: mudrov_viktor@mail.ru

Anastasiya Yu. Alekseyeva, MD;
ORCID: <https://orcid.org/0000-0001-5061-8026>;
e-mail: mironenkoanastasia4@gmail.ru

ОБ АВТОРАХ

Елена Владимировна Фалько;
e-mail: p-2000f@yandex.ru

Виктор Андреевич Мудров, канд. мед наук;
ORCID: <https://orcid.org/0000-0002-5961-5400>;
e-mail: mudrov_viktor@mail.ru

Анастасия Юрьевна Алексеева;
ORCID: <https://orcid.org/0000-0001-5061-8026>;
e-mail: mironenkoanastasia4@gmail.ru