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Intrauterine Fetal Demise Secondary to Furcate Insertion of the Umbilical Cord. Case Report and Literature Review

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Abstract:

Furcate umbilical cord insertion is a rare anomaly (0.1% incidence). It is considered an identifiable cause for fetal demise, taking into consideration that all unexplainable causes constitute 76% of all fetal demises, this is an identifiable cause associated with a placental alteration. Our objective is to document a case of fetal demise secondary to furcate umbilical cord insertion and review related literature. The case presented is of a 29-year-old woman, undergoing her first pregnancy of 40.3 weeks. She reports diminished fetal movements, ultrasound findings confirm fetal demise. Pregnancy is interrupted, placenta and membranes were sent for pathology evaluation, reporting furcate cord insertion. Furcate umbilical cord insertion is a rare anomaly associated to perinatal death. Diagnosis should be confirmed by histopathology. Diminished fetal movements is an important clinical sign, indicating fetal compromise. This is a case of fetal demise associated to placental insufficiency and not a traumatic event as reported in other publications. An ultrasonographic evaluation in a suspicious case, should make a thorough evaluation of the cord to evaluate these malformations.

Introduction

Intrauterine fetal death (IFD) has a worldwide incidence of 2.6 million cases each year (from 28 weeks of gestation, according to the World Health Organization (WHO)), or 18.4 per 1,000 births **[**1**]**. Risk factors associated with IFD include maternal obesity, advanced maternal age, smoking, decreased fetal movement, intrauterine growth restriction, and hypertensive disorders of pregnancy. Abnormalities in placental structure and function can also be determining factors for IFD **[**2**]**.

In developed countries, cases of IFD usually occur in the antepartum period, whilst in developing countries, about 50% of cases occur during labor **[**2**]**. Pathologies of the placenta and umbilical cord contribute to 20% of the causes of IFD **[**3,4**]**, these can be divided into 6 groups; two groups can initially be diagnosed prenatally: 1) macroscopic lesions of the placenta, membranes, and umbilical cord, 2) complications of monochorionic placentation, 3) abnormal development of the villous parenchyma, 4) infectious diseases, 5) compromised maternal and/or fetal circulation, 6) hematological alterations dependent of an histological evaluation **[**5**]**.

Furcate insertion of the umbilical cord (FI) is among the macroscopic lesions of the placenta and umbilical cord, being a rare entity and with an approximate prevalence of 0.1% in all pregnancies. FI is defined as a separation or branching of the umbilical vessels before their insertion into the placenta or adjacent membranes. Close to the cord insertion site, furcated vessels lack surrounding Wharton's jelly and, therefore, by losing this protection, they are more exposed to injury, aneurysmal changes, thrombosis, vessel rupture, hemorrhage, and IFD **[**6**]**.

Among the reported cases, spontaneous rupture of the umbilical vessels and fetal death in term pregnancies have been described, although this involves trauma, which, from a legal point of view, is considered a natural death or death associated with the malformation **[**7**]**. Likewise, there are several reports where the presence of FI is associated with umbilical cord cysts, varicose veins, thrombosis, single umbilical artery, velamentous and marginal insertion, intrauterine growth restriction and fetal malformations **[**7-10**]**.

FI cases appear to show normal cord insertion into the central part of the placenta; therefore, prenatal diagnosis can be difficult during routine ultrasound **[**11**]**. According to the literature, only three cases of prenatal suspicion confirmed by FI pathology have been reported **[**6,9,11**]**. The first article published in 2015 describes the FI as part of a double placenta with a bifurcated umbilical cord **[**9**]**, while the second article, published in 2017 describes an umbilical cord that contained three vessels inserted in the center of the placenta, but the umbilical vessels were separated from the cord's Wharton jelly prior to its insertion into the placenta **[**11**]**. A third article published in 2020 describes a bifurcation of the umbilical vein with a branch to the placenta and another to the adjacent membranes **[**6**]**. The diagnosis in the last two cases was carried out at 33 and 35 weeks of gestation, respectively, and in all three the pregnancies were terminated at 37 weeks of gestation without subsequent complications.

Clinical case

We present the case of a 29-year-old patient, primiparous, undergoing a normal evolutionary pregnancy of 40.3 weeks of gestation by date of last menstrual period. Relevant patient's history is diagnosis of hypothyroidism established at age 16, under control with hormone replacement therapy with levothyroxine, 50 micrograms every 24 hours, 6 times a week. First and second trimester ultrasonographic markers were within normal parameters, as well as third-trimester growth ultrasound at 34 weeks of gestation, which reports a posterior placenta grade III on the Grannum scale, with no data of hematoma or detachment, implanted at a normal distance from the internal cervical oz. Rest of her background irrelevant to the case.

The patient referred decreased fetal movements upon awakening, she contacted her physician who performed an obstetric ultrasound, finding absence of cardiac activity. Abdominal resolution of the pregnancy was decided since there were no cervical conditions for the vaginal route. A lifeless, female product of 3325 grams was obtained, amniotic fluid with meconium +++, no evidence of secondary bleeding from umbilical cord trauma or placental abruption. The placenta and annexes are sent for study by microbiology, which did not report bacterial development and ruled out any infectious process. The histopathological study reports: a) placenta weighing 456 grams (weight in the 10th percentile), b) thin, hyper-coiled cord, with a diameter of 0.8 cm (diameter less than the 10th percentile), with FI, and squamous metaplasia of the amniotic epithelium (see Figure 1 and 2), c) delayed villous maturation in 30% of the examined surface, d) multifocal chorangiosis, e) fibrosclerosis in the vasculature of the chorionic plate, f) chronic chorioamnioitis with subacute meconium/ chronic, g) vascular ectasia of the chorionic plate, h) extensive accentuated vascular congestion, i) mixed degeneration with cysts in fetal membranes and decidual lamellar infarcts. Prior to this, all his ultrasonographic studies were reported as normal and without alterations.

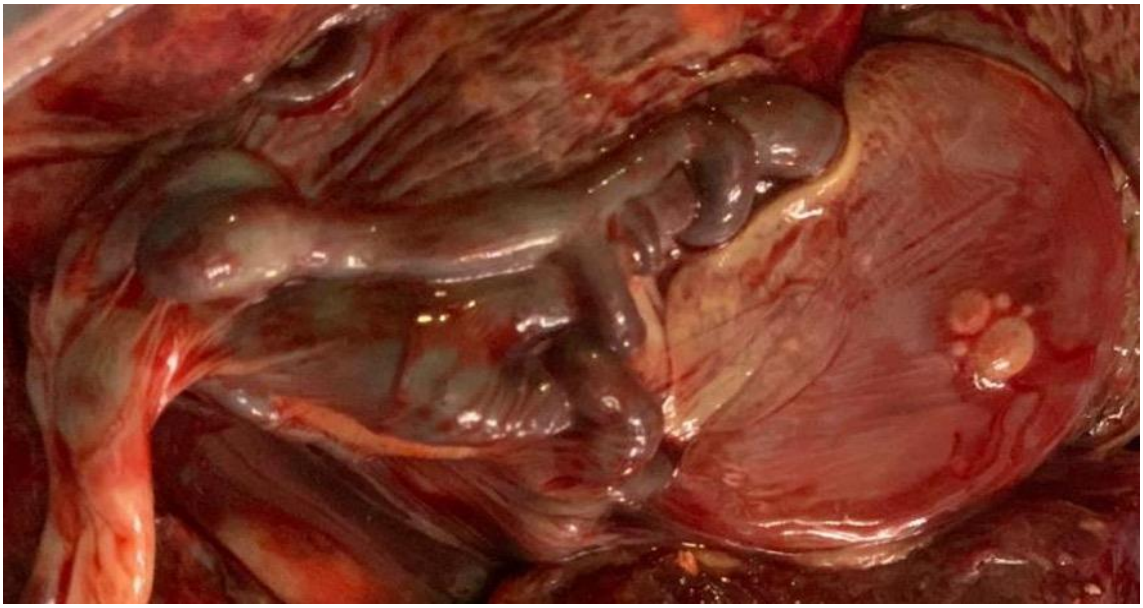

Figure 1. Furcate insertion of umbilical vessels to the fetal placental face, in this image we can see the bifurcation of the umbilical vent and the umbilical arteries, prior to their insertion into the placenta.

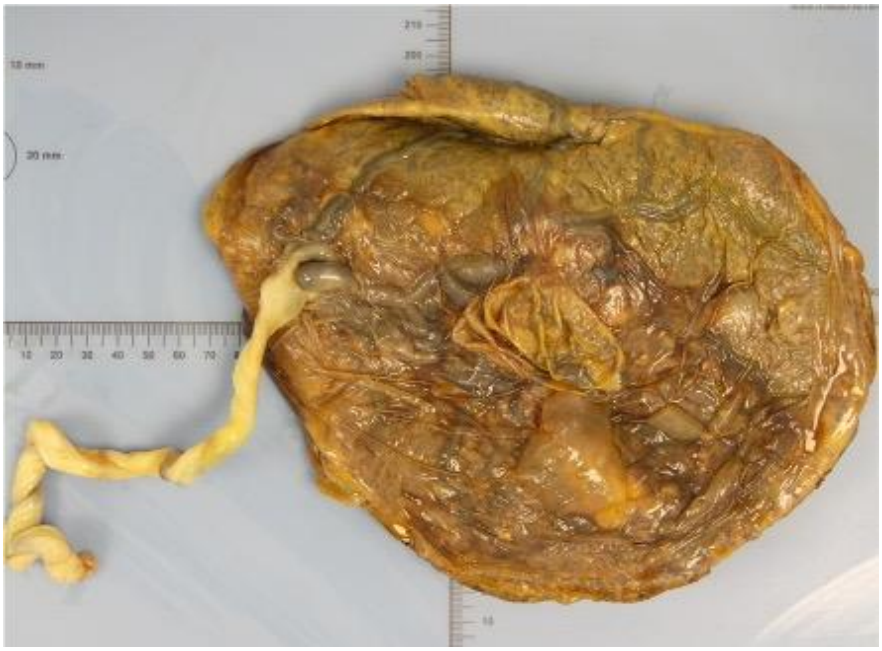

Figure 2. In this image the full size of the placenta can be appreciated with the alterations in the fetal face related to the furcate insertion of the umbilical vessels.

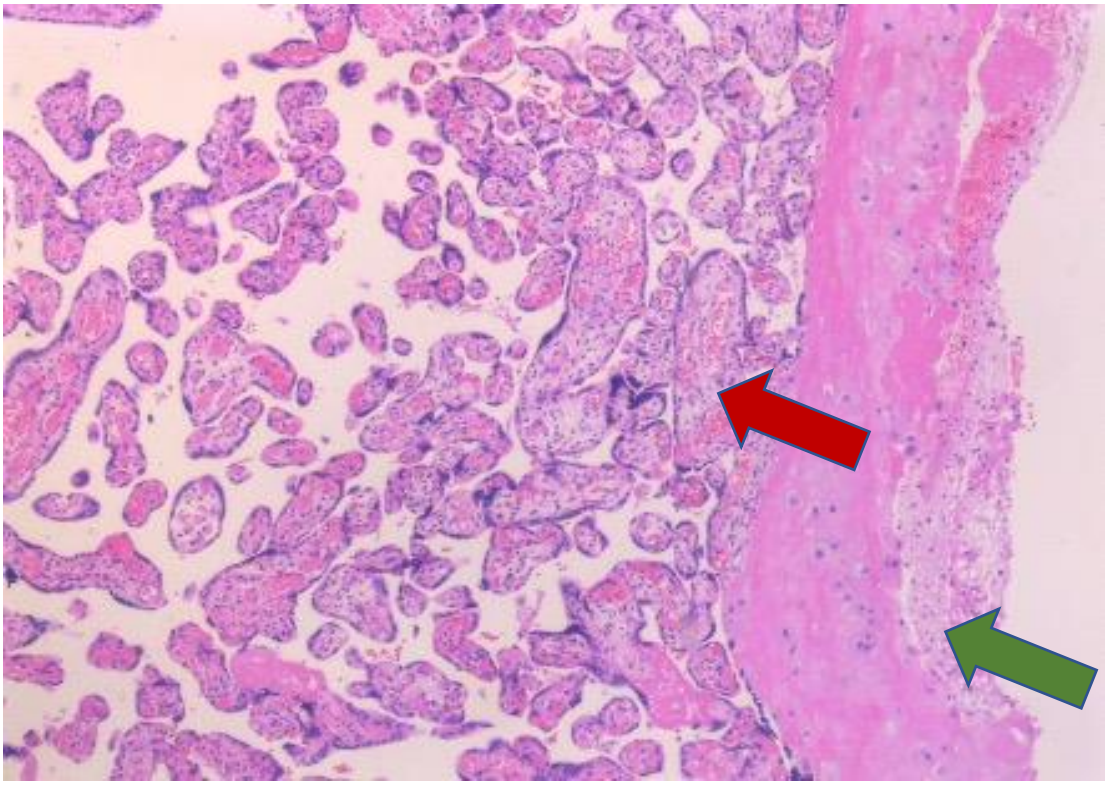

Figure 3. Histological section of the placenta showing myxoid degeneration (red) and lamellar infarcts (green).

Discussion

FI of the umbilical cord is a rare malformation, the diagnosis of which is mainly histopathological and has been associated with adverse perinatal outcomes. A review carried out by Kryzanowski et al combined this entity with velamentous insertions of the umbilical cord, mentioning an incidence of 1% [12], however, when isolated, Kosian et al reports an incidence of barely 0.1% [13]. To date, the literature does not report a percentage of prenatal diagnosis for FI of the umbilical cord. This structure is visible from 42 days of gestation. During first trimester it is recommended to observe the insertion on the placenta, since visualization is difficult in advanced gestational stages or in the case of placentas with posterior insertion. Its location is usually aided with the use of "color Doppler" mode in order to follow the cases of chorionic plaque. If it is not possible to assess the placental insertion, a tangential visualization of the placental surface can be performed to observe the vessels by "color Doppler" while approaching the umbilical cord insertion [12,14]. It is considered that ultrasound in the second trimester with an experienced operator and state-of-the-art ultrasound equipment would have the capacity to detect these alterations, although it is a rare alteration, and its prenatal detection capacity has not been reported. Verifying the presence of the three vessels and the greatest possible extension of the cord may not be enough. In addition, in several cases the position, presence of other structures and other situations complicate the complete visualization of the cord, as in the case presented [13].

To date, few cases of FI have been described, which is why the risks associated with this alteration are poorly understood. However, there are associations between FI and IFD, fetal hemorrhage, and intrauterine growth restriction [13].In a review of 132 cases of FI, 56% (75 patients) had perinatal complications. Among them, a "non-reassuring" cardiotocographic record is described in 26.5%, admission to neonatal intensive care 17.6%, meconium-stained amniotic fluid 16.3%, retained placenta 5.1%, placenta previa 3.1%, shoulder dystocia 2%, uterine rupture 2%, uterine atony 2%, intra-amniotic infection syndrome 1%, and abruptio placenta normoinserta 1% [13].

In a case reported by Cohen et al the patient went for evaluation due to decreased fetal movements, and upon reviewing the case, found no evidence of hemorrhage, both situations like our case. The authors report that most cases of intrauterine fetal death associated with umbilical cord FI have no evidence of trauma, so the true mechanism of death is still unknown [6].

We do not know whether there is a direct association between meconium-stained amniotic fluid and FI, however, we consider that it probably occurred secondary to fetal distress. Considering FI as a vascular disorder that is exposed to partial or total occlusion, in partial cases, vascular stasis, thrombosis, fibrin deposition in large vessels of the chorionic plate and villus stalk; progression of

these pathologies can lead to the presence of hyalinized avascular villi; hence the accumulation of these alterations generates hypoperfusion. Taking this into account, the presence of meconium-stained amniotic fluid is secondary to a vagal response in response to stress (seen in 30% of vaginal deliveries) **[**15**]**. It would not be difficult to associate this vascular alteration with a condition of hypoperfusion and its associated morbidities.

Decreased fetal movements are associated with IFD and other adverse perinatal outcomes such as fetal-maternal hemorrhage, umbilical cord accidents, oligohydramnios, and long-term neurodevelopmental disability. The hypothesis is that this decrease in fetal movements is due to a compensatory response in which there is placental insufficiency, allowing the fetus to conserve energy, which is an early warning sign of a deterioration in fetal condition **[**16**]**.

During prenatal control, "color Doppler" ultrasound should be used to confirm the insertion site of the umbilical cord in the placental parenchyma, as well as its location. If FI or any other alteration in the structure of the cord is suspected, it is necessary to take precautions against sudden IFD and explain to patients the warning sings, risks, and associated complications.

In most cases, there are favorable obstetric results, so umbilical cord FI continues to be overlooked, mainly due to lack of knowledge of this abnormal insertion **[**13**]**.

In case this alteration is detected by prenatal ultrasound, it is recommended labor induction at 37 weeks of gestation to avoid any complications, and in the case of additional findings such as umbilical cord cysts, ectasia or thrombosis, interruption at earlier gestational ages is advised **[**13**]**.

Although the etiology of FI is uncertain, some findings have been described, such as fibrin deposits or areas of microinfarcts, which could suggest that the initial insertion of the cord was normal and that there was later degeneration of the placental tissue below the cord. This could result in the formation of cystic lesions, causing the umbilical vessels to separate from the placental parenchyma **[**11**]**. In our case, the data reported by pathology such as delayed villous maturation, fibrosclerosis, myxoid degeneration, and laminar infarcts (Figure 3) would support this theory. The presence of chorangiosis is also described, defined as villous hypervascularity characterized by more than 10 capillaries in more than 10 chorionic villi in different areas of the placenta. Normally, the villi contain less than 5 capillaries **[**16**]**. This can be associated with greater neonatal morbidity and mortality, and its prevalence increases in cases when there are placental lesions that cause chronic hypoperfusion. An association between chorangiosis and placental pathological conditions has been observed, such as placental abruption, villitis and umbilical cord abnormalities **[**17**]**.

Hypothyroidism, as an independent factor associated with placental insufficiency, could be based on trophoblastic invasion of the decidua and spiral arteries, since these require thyroid hormone for adequate placentation. Thyroid hormone regulates the secretion of various factors and cytokines critical to this process [18]. Hypothyroidism, among other perinatal complications, is associated with IFD and this is possibly due to placental hypoplasia together with other placental abnormalities [19]. Our patient did not present alterations in her metabolic control and her control studies were always normal.

Essential components of IFD evaluation include histopathologic examination of the placenta, umbilical cord, and fetal membranes, and ideally fetal biopsy and genetic studies [20].

Conclusion

FI of the umbilical cord is an anomaly with a low incidence and difficult to diagnose prenatally, since in most cases it has been described as an incidental finding, as in our case. Histological and structural alterations of the placenta can compromise vascular perfusion, increasing the risk of IFD and endorses the need for prenatal ultrasonographic studies to be performed by Maternal Fetal Medicine specialists who, in case of having any risk factor or suspicion of these alterations, additional studies may be required. Our case supports the concept that an alteration in placental perfusion may be the cause of IFD.

Hypothyroidism, although not a described risk factor, can be considered as a risk cofactor for a placenta with FI of the cord.

The decrease in fetal movements is an important obstetric alarm data due to its association with adverse perinatal outcomes.

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