Uterine Rupture in A Pregnancy Complicated By A Giant Placental Chorioangioma: Case Report

Dev Bir Plasental Koryoanjiyoma ile Komplike Olan Bir Gebelikte Uterin Rüptür

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ABSTRACT No case of uterine rupture associated with a chorioangioma has been reported up to date. Herein, we present a pregnancy complicated with spontaneous uterine rupture due to polyhydramnios secondary to a giant placental chorioangioma in a patient with two previous cesarean sections at the early third trimester. The patient who is at 29 weeks of gestation was admitted because of sudden abdominal pain and distension. Her obstetric history revealed two uncomplicated previous lower segment cesarean sections. The prenatal course of this pregnancy had been uneventful so far and her only ultrasonographic evaluation which was performed three weeks ago was reported to be normal. Upon vaginal examination bleeding was detected and the uterine cervix was not dilated. Ultrasound revealed a single live fetus with a transverse lie and a markedly increased amniotic fluid volume and the examination did not reveal any obvious fetal abnormality; however the placenta showed a well-defined solid mass and an amniotic sac protruding out from the Kerr incision was seen. On cardiotocography, severe persistent fetal bradycardia was detected. An emergency cesarean section was performed with the prediagnosis of uterine rupture secondary to polyhydramnios due to a possible placental chorioangioma. Final pathology was chorioangioma

Key Words: Uterine rupture; polyhydramnios; ultrasonography; diagnosis; chorioangioma

ÖZET Bugüne dek, koryoanjiyoma ile ilişkili uterin rüptür olgusu bildirilmemiştir. Burada, üçüncü trimesterin erken döneminde, dev bir plasental koryoanjiyomaya sekonder olarak gelişmiş polihidramniosa bağlı spontan uterin rüptürle komplike olan bir gebelik olgusu sunmaktayız. Yirmi dokuzuncu gebelik haftasında olan hasta, ani başlayan karın ağrısı ve karında şişkinlik şikayetleri ile başvurmuştur. Obstetrik hikâyesinde iki adet geçirilmiş sezaryen bulunmaktadır. Gebeliği bugüne dek sorunsuz geçmiş ve üç hafta önce yapılan ultrasonografinin normal olduğu bildirilmiştir. Vajinal muayenede kanama vardı ve serviks dilate değildir. Ultrasonda transvers duruşta tek bir canlı fetus ve belirgin olarak artmış amniyotik sıvı hacmi saptanmıştı, görünür fetal anomali yoktu; ancak plasentada solid bir kitle ve Kerr insizyon hattından taşan amniyotik kese görülmekteydi. Kardiyotokografide belirgin kalıcı fetal taşikardi vardı. Hastaya, olası plasental koryoanjiyomaya sekonder acil sezaryen yapılmıştır. Patolojik inceleme tanıyı doğrulamıştır.

Anahtar Kelimeler: Uterin rüptür; polihidramnios; ultrasonografi; tanı; koranjiyom

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Chorioangiomas are the most common benign placental tumors with a reported incidence of 1%.[1] Larger tumors measuring more than 4 cm are rarely seen in obstetric practice, but they are associated with serious maternal or fetal complications.[1-3] These complications include polyhydramnios, preterm delivery, nonimmune hydrops, and fetal heart failure.[1-3] Among these, polyhydramnios is one of the major risk factors which

may give rise to premature labor. Furthermore, polyhydramnios may also potentially result in uterine rupture especially in a scarred uterus due to overdistention. However, no case of uterine rupture secondary to a placental chorioangioma has been reported up to date when we made a MEDLINE search with the keywords "uterine rupture, polyhydramnios and chorioangioma". Uterine rupture which is a major obstetric complication most commonly involves a previous uterine scar and the rupture may be either traumatic or spontaneous.[4,5]

Herein, we present a pregnancy complicated with spontaneous uterine rupture due to polyhydramnios secondary to an unrecognized giant placental chorioangioma in a patient with two previous cesarean sections at the early third trimester.

CASE REPORT

A 24-year-old woman in her third pregnancy was admitted to our emergency department at 29 weeks of gestation because of sudden abdominal pain and distension. Her obstetric history revealed two uncomplicated previous lower segment cesarean sections; the first one performed for a demised fetus with breech presentation two years ago and the second for previous cesarean section a year ago. She did not receive routine antenatal care but the prenatal course of this pregnancy had been uneventful so far and her only ultrasonographic evaluation which was performed three weeks ago was reported to be normal. Upon vaginal examination bleeding was detected and the uterine cervix was not dilated. On the other hand, the patient was hemodynamically stable.

Ultrasound examination revealed a single live fetus with a transverse lie and a markedly increased amniotic fluid volume. Fetal growth parameters corresponded with period of gestation with an amniotic fluid index of 31. The examination did not reveal any obvious fetal abnormality; however the placenta showed a well-defined solid mass measuring 78 ×73 mm. The mass was predominantly hypoechoic with a single hyperechogenic line in the middle and tiny calcifications at the periphery and could be easily distinguished from the normal placental tissue (Figure 1). Furthermore, a 14 × 13 cm amniotic sac protruding out from the Kerr incision was seen (Figure 2). On cardiotocography, severe persistent fetal bradycardia (61 bpm) was detected. An emergency cesarean section was performed with the prediagnosis of uterine rupture secondary to polyhydramnios due to a possible placental chorioangioma. Laparotomy confirmed rupture of the uterus with extrusion of the placenta and fetal extremities into the upper abdomen. A female infant weighing 1130 g was born with Apgar scores of 0 and 2 after 1 and 5 min, respectively, and she died the following day at the neonatal intensive care unit. There was a large placental mass with a diameter of approximately 10 × 9 cm at the fetal side, which detached from the placenta spontaneously (Figure 3).

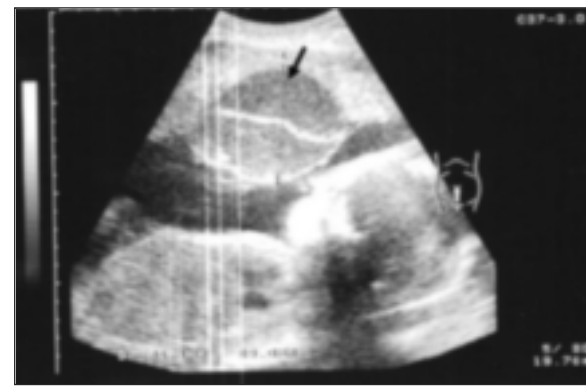

FIGURE 1: Ultrasound image showing the placental chorioangioma. The arrow points the chorioangioma.

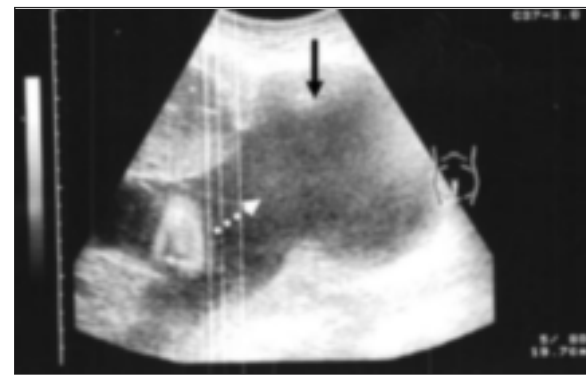

FIGURE 2: Ultrasound image showing the amniotic sac protruding out from the Kerr insicion dehiscence.
The black arrow points the amniotic sac protruding from the previous cesarean scar.
The white dashed arrow points the scar dehiscence.

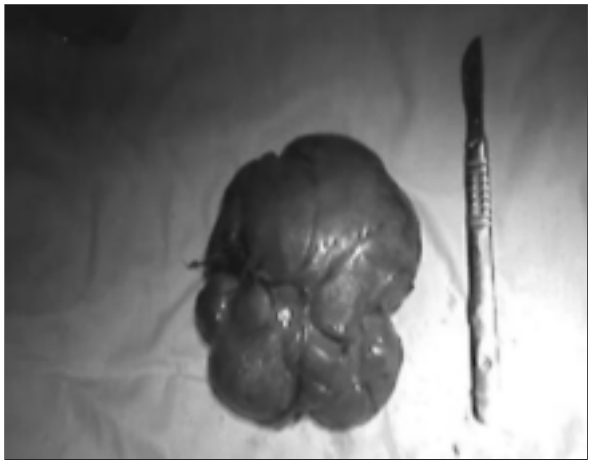

FIGURE 3: The macroscopic view of the chorioangioma detached from the placenta.

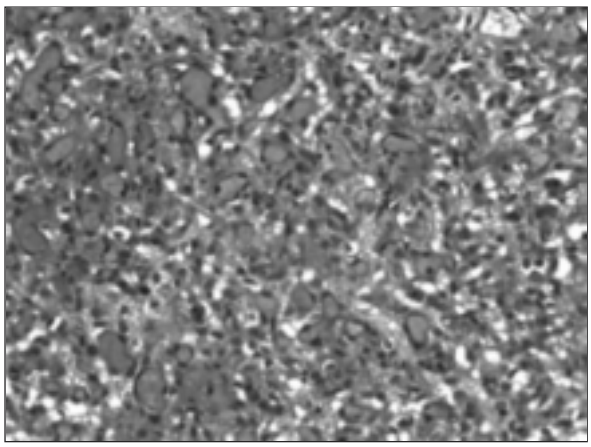

FIGURE 4: H&E section of the chorioangioma (x400) showing expanded vascularity and increased stromal cellularity.

Final pathology revealed a large 10 cm subchorionic single nodular mass on the cut surface of the placenta, consistent with a chorioangioma. Microscopically, the tumor was composed of a network of proliferating capillaries. Mitotic figures were easily detected and the nuclei were small, uniform and lacked atypia. Hematoxylin & eosin staining (Merck®, Bio optica®) showed expanded vascularity and increased stromal cellularity (Figure 4). Immunohistochemical study using CD 34 (DakoCytomation®) antibody was also carried out in order to confirm the presence of CD 34 antigen on immature hematopoietic precursor cells.

DISCUSSION

Most of the chorioangiomas that are recognized histologically are small and remain clinically insignificant until they exceed 5 cm in diameter.[2] The incidence of larger chorioangiomas is very low, approximately 1/9000.[6] The chorioangioma in the current study is one of the largest reported in the literature. In a large clinical study, He et al described 12 placental chorioangioma cases, including one that reached 11 cm in size and three reports were published which described a chorioangioma that reached 12 cm in size; while the other studies have reported chorioangiomas between 5-8 cm in size.[7-10] Maternal or fetal complications are predominantly associated with large chorioangiomas and the overall perinatal mortality rate associated with them approaches 30%.[1,11] Besides, it is well known that, the larger the size of the chorioangioma, the greater the chances of complications and adverse fetal outcomes.[12]

Although the underlying pathophysiology for these complications is not fully understood, a prominent role for arteriovenous shunting and sequestration of red blood cells and platelets by the tumor has been postulated.[1-3] Prematurity related to polyhydramnios and an association with disseminated fetal hemangiomas are other causes of perinatal mortality.[11]

In this case report, we aimed to present a case of spontaneous uterine rupture which developed due to polyhydramnios secondary to a giant chorioangioma 10 cm in diameter. This seems to be the first reported case of uterine rupture in the literature secondary to a placental chorioangioma.

The suggested mechanism for evolution of polyhydramnios in cases with placental chorioangioma include the transudation of fluid from the tumor into the amniotic fluid, compression of the umblical vessels by the tumor and increased secretion of fetal metabolites through the tumor.[13] In our case, the ultrasound examination revealed a a markedly increased amniotic fluid volume associated with a large chorioangioma. Similarly, Prapas et al also reported that six out of their seven cases with chorioangioma demonstrated polyhydramnios in the late second or third trimester.[14] In other studies, it was found that polyhydramnios correlated with tumor size and occured in 18-35% of cases with chorioangiomas.[2]

Uterine rupture is a major obstetric hazard, most commonly involving a previous uterine scar.[4,5] In our case, the rupture seems to have developed spontaneously due to the overdistension of the uterus secondary to polyhydramnios caused by the chorioangioma before the onset of uterine contractions. Since the patient did not receive any antenatal care, the unawareness of both polyhydramnios and the chorioangioma possibly have lead to spontaneous rupture of the scarred uterus. Interestingly, the fetus in our case showed no signs of fetal heart failure, nonimmune hydrops or fetal growth restriction and the only complication caused by such a large chorioangioma was polyhydramnios and uterine rupture as the consequences. Although giant chorioangiomas usually lead to serious fetomaternal complications, they may sometimes remain asymptomatic.[15,16]

Besides, the large placental chorioangioma described in the current study was not identified on ultrasonography at 26 weeks. There seems to be two possibilities for undetecting the tumor on that previous examination: the first one may be inadequate examination of the placenta and the second may be that rapid lesion growth did not occur until later in gestation and thus, the lesion was significantly smaller at 26 weeks. Since chorioangiomas may have very rapid proliferation rates, a larger lesion may have initially been too small to be detected.

An earlier diagnosis of the chorioangioma might have improved the prognosis of this case by providing an opportunity for treatment of polyhydramnios and for close maternal / fetal surveillance. Therefore, examination of the placenta and the amniotic fluid is as far important as evaluation of the fetal growth and anatomy during routine obstetric ultrasonography.

Obstetric sonography is the mainstay in diagnosing chorioangioma, which appears on gray-scale sonography as a hypoechoic mass bulging on the fetal surface of the placenta in most cases. Calcification and necrosis may also be observed. In our case, the chorioangioma was a well-circumscribed hypoechoic lesion with a central hyperechogenic line in the middle (the feeding vessel) and peripheral calcifications. However, chorioangiomas have similar sonographic appearance to placental hematomas, placental teratoma, and degenerating fibroid, but the use of color Doppler imaging can facilitate the diagnosis if vessels are identified within the tumor.[17] We could not use the Doppler in our case due to unavailability of the equipment during the night shift. Although the calcifications in chorioangioma has previously been reported to be associated with reduced blood flow in the tumor and a favorable outcome, despite having calcifications the chorioangioma in this case had resulted in a catastrophic outcome.[18,19]

As a conclusion, placental evaluation should always be performed during routine obstetric ultrasonography. Although rarely seen, examination of the placenta enables detection of large chorioangiomas and other placental tumors. Consequently, this might improve the prognosis of the pregnancy by close surveillance and prompt clinical management of the associated complications.

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