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Antenatal embolization of a large chorioangioma by percutaneous Glubran 2 injection

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ABSTRACT

We describe a case of a large chorioangioma diagnosed at 18 weeks' gestation. Because of advanced fetal heart failure at 23 weeks' gestation, embolization of the chorioangioma's vessels was performed by percutaneous injection of Glubran 2 surgical glue. There was no immediate secondary effect of treatment. Devascularization was complete and durable. Signs of fetal cardiac failure normalized after 1 month and a healthy infant was delivered at 38 weeks. To our knowledge this is the first reported case of perinatal survival after successful embolization of a chorioangioma using tissue glue. Copyright © 2010 ISUOG. Published by John Wiley & Sons, Ltd.

CASE REPORT

A 27-year-old primigravida was referred at 22 weeks' gestation for suspicion of fetal anemia. Her medical and familial histories were unremarkable. The pregnancy had been uneventful until 18 weeks, when the alpha-fetoprotein level measured in the patient's serum for Down syndrome screening was 9.4 multiples of the median (MoM) for gestational age. At this point ultrasound examination detected a placental mass of 4 cm in diameter; fetal morphology was normal. At 22 weeks, ultrasound examination (Voluson E8 Expert, GE Healthcare Ultrasound, Milwaukee, WI, USA) showed a eutrophic fetus with cardiomegaly (cardiothoracic index 0.65), pericardial effusion, thick cardiac walls and large (6.2 mm) and pulsatile umbilical vein. The amniotic fluid index was normal. Maximal systolic velocity of the middle cerebral artery exceeded 1.7 MoM (44 cm/s). The placental mass that had been previously detected presented as a $7 \times 7 \times 8$-cm highly vascularized solid tumor arising from the upper part of the placenta and protruding into the amniotic cavity (Figure 1). A diagnosis of fetal heart failure due to a large chorioangioma was highly suspected. Cordocentesis showed a hemoglobin concentration of 6.9 g/dL (hematocrit 21%) and platelet count of 76 000/μL; karyotype was normal, 46,XX.

Two *in-utero* transfusions were performed over a 4-day period because of a rapid fall in the level of fetal hemoglobin. Magnetic resonance imaging examination confirmed the diagnosis of chorioangioma (Figure 2). After extensive counseling the couple agreed to percutaneous embolization of the tumor. The procedure was performed under local anesthesia and ultrasound guidance at 23 weeks' gestation and took 15 min. A 22-gauge spinal needle was inserted into the arterial feeding vessel within the tumor, flushed with 5 mL of 5% dextrose solution followed by injection of 6 mL mix of Glubran 2 (GEM Srl, Viareggio, Italy) diluted with Lipiodol Ultra-Fluide (Guerbet, Villepinte, France) at a ratio of 1/5 following the recommendation of our interventional radiologist. Immediate interruption of the vascular supply to the tumor was observed by grayscale ultrasound and confirmed by color Doppler imaging (Figure 3).

Follow-up confirmed durability of the devascularization of the chorioangioma. The Doppler signal of the umbilical vein and its diameter normalized rapidly and the velocity of the middle cerebral artery remained normal, but the cardiothoracic index and hypertrophic appearance of the cardiac walls normalized only after 1 month. At 32 weeks, fetal biometry indicated intrauterine growth restriction, with all other parameters of fetal well-being remaining normal. Doppler imaging showed a protodiastolic notch in the two uterine arteries. A female neonate was delivered at 38 weeks by Cesarean section for abnormal umbilical artery resistance index and breech presentation. Her birth

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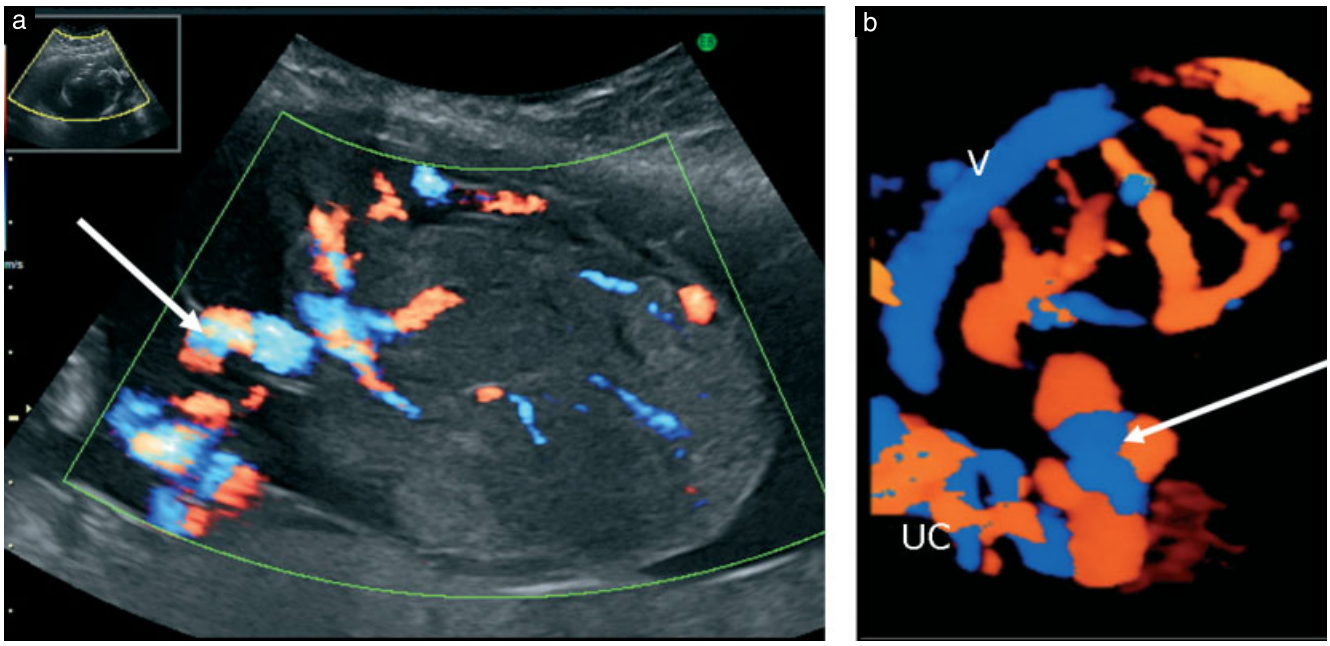

Figure 1 (a) Doppler ultrasound image of the chorioangioma before embolization showing hypervascularization and two feeding vessels (arrow), one of which had arterial pulsatility at the same rate as the fetal heart; the other had a venous spectrum. The mass was surrounded by a thin membrane. (b) Three-dimensional color Doppler image of the mass showing the origin of the feeding vessels (arrow) of the chorioangioma close to the placental umbilical cord insertion (UC). A large vein (V) ran superficially around the mass.

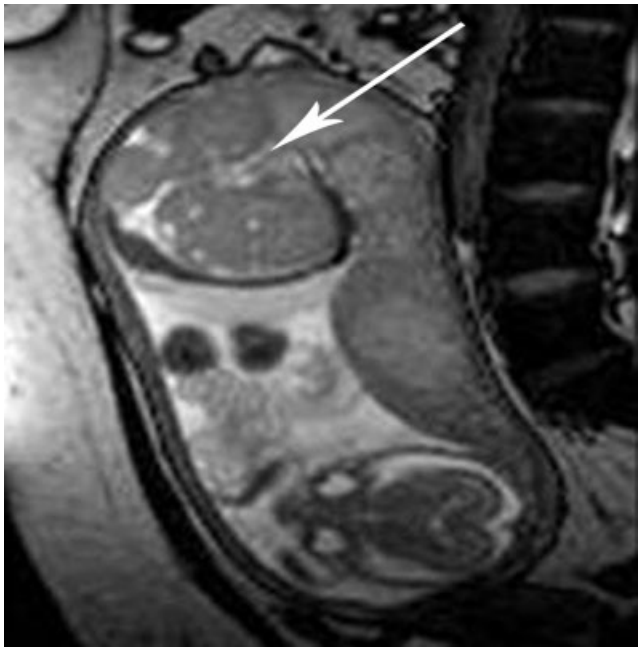

Figure 2 Sagittal T2-weighted magnetic resonance image of the uterus showing a fundal mass presenting a similar signal to that of the adjacent placenta, indicating its origin (arrow).

weight was 2600 g and Apgar scores were 8, 9 and 10 at 1, 5 and 10 min, respectively. The placenta weighed 535 g (median for age, 467 g). Gross examination of the placenta revealed a necrotic mass of 8 × 6 × 4 cm connected to the placenta by two vessels. Histological examination confirmed the diagnosis of chorioangioma with necrotic changes, and embolization material was observed in the vessels (Figure 4).

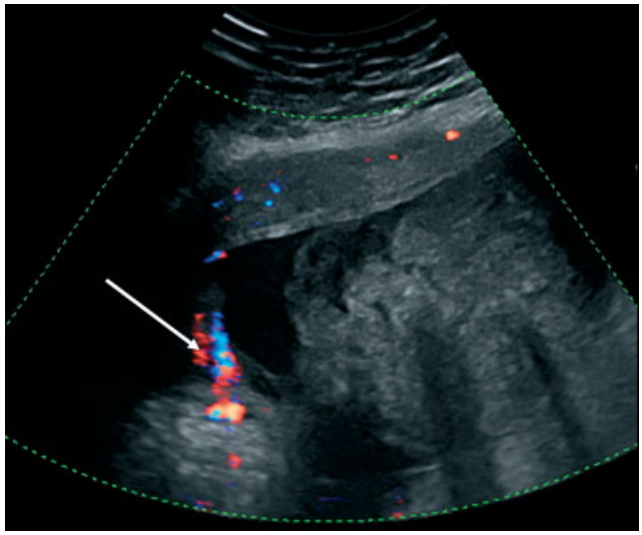

Figure 3 Color Doppler ultrasound image of the chorioangioma after devascularization, showing a lack of blood flow in the vessels of the chorioangioma (arrow shows the placental umbilical cord insertion with blood flow present).

DISCUSSION

Chorioangioma is the most common benign tumor of the placenta, with an overall prevalence of 0.9% of pregnancies; however more than 50% are only visible by careful dissection of the placenta[1–5]. The incidence of large chorioangioma (> 4 cm) is low, ranging from 1 in 9000 to 1 in 50 000 pregnancies[1,2].

About half of large chorioangiomas are associated with fetal and/or maternal complications[1]. Principal fetal complications are hemolytic anemia, thrombocytopenia, growth restriction, congestive heart failure, non-immune

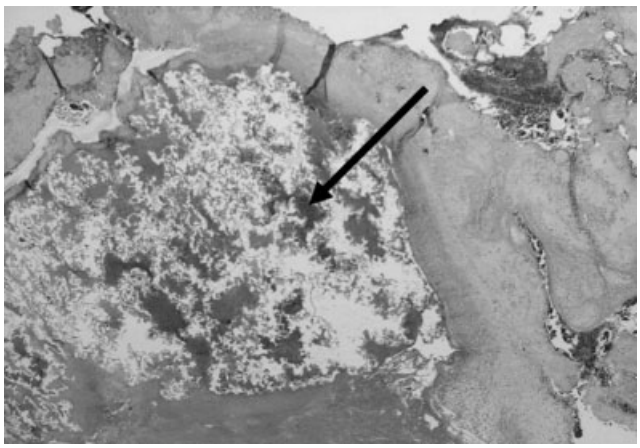

Figure 4 Histological examination of the vessels of the chorioangioma showed the presence of embolization material (arrow) (H&E, original magnification × 20).

hydrops and fetal demise; polyhydramnios and preterm labor are common. The mother may develop hemolytic anemia, thrombocytopenia, disseminated intravascular coagulation, pregnancy-induced hypertension or antepartum hemorrhage[1–4]. The overall perinatal mortality is about 30–40%[5,6]. As in our case these complications may be acute and chorioangiomas may grow rapidly, requiring close monitoring of the pregnancy[1,5,7]. In this case the fetus developed anemia, moderate thrombocytopenia and cardiac failure, but no polyhydramnios was observed.

Demonstration of blood flow in the mass by color Doppler ultrasound is useful for differential diagnosis with non-vascular placental tumors although not all chorioangiomas are vascularized[1–4]. High vascularization of chorioangioma observed on color Doppler is now recognized as an independent factor of a more severe pattern of fetal complications. These chorioangiomas present more frequently with elevated maternal serum alpha-fetoprotein, as in our case[3]. Color Doppler is necessary to study the exact location of the chorioangioma's feeding vessels before attempting *in-utero* devascularization and to monitor its effectiveness[8]. The potential value of repeated three-dimensional power Doppler evaluations in the assessment of the hemodynamics and prognosis of chorioangiomas has previously been suggested[4].

When complications appear, different treatments are possible if gestational age precludes delivery[5,6]. An extensive review of invasive treatment options was recently published by Sepulveda *et al.*[6]. Treatment by intrauterine blood transfusion or amniodrainage has been attempted[7], although the aim of intervention should be to block the vascular supply of the tumor to permit the fetus to recover and prevent further complications[6]. The most frequently used methods are: endoscopic laser coagulation (five cases, two survivors)[6,8]; absolute alcohol injection (six cases, four survivors, two re-interventions)[6]; and interstitial laser coagulation (four cases, all survived, two re-interventions)[5,6]. Other procedures are endoscopic suture ligation[7] and embolization by microcoil[9] or enbucrilate injection[10] (no survivor). The choice of method depends on the diameter and location – superficial or deep – of the feeding vessels and the distance between umbilical cord and chorioangioma[5–8]. Poor pregnancy outcome after endoscopic laser therapy can be expected when a feeding vessel is deep and/or large and with close insertion of the umbilical cord[6,8].

The present study reports the successful use of tissue glue to treat a chorioangioma with a large feeding artery (7.8 mm) that was located close to the umbilical cord insertion. This simple technique requires only local anesthesia, but precise localization of the needle in the feeding vessel is necessary. The potential advantages of using tissue glue have already been described[10]. Glubran 2 is a new cyanoacrylate tissue glue widely used in interventional radiology. The first successful devascularization of a chorioangioma using tissue glue was reported in 2005, however, due to premature labor soon after the procedure the baby died at 26 weeks' gestation[10]. Our case emphasizes the potential of tissue-glue injection, which remains a less invasive *in-utero* procedure. It can be an alternative to the use of endoscopic or interstitial laser according to the location of the chorioangioma's feeding vessels and local skills.

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