



Exploring the Prospects of Antenatal Microcoil Embolization of Giant Placental Chorioangioma: Case Report and Review of Literature

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Abstract

Giant placental chorioangiomas, which measure more than 4 cm, are associated with adverse maternal, fetal, and neonatal outcomes, with an estimated perinatal mortality of 30 to 40%. Early diagnosis, close prenatal surveillance, intrauterine intervention, and timely delivery would play a vital role in improving pregnancy outcomes. Using conventional treatment options like amniodrainage to reduce intrauterine pressure and intrauterine blood transfusion to correct fetal anemia fails to tackle the underlying pathophysiology. Interventions aimed at devascularizing the tumor, such as injection of alcohol or surgical glue, use of bipolar cautery, placement of vascular clips, interstitial laser, and radiofrequency ablation, have had mixed results due to their invasive nature and associated complications. In this case report, we discuss a minimally invasive, percutaneous, extraamniotic method of focal occlusion accomplished by microcoil embolization of the feeding vessel. The combination of slow blood flow and endothelial vessel damage due to the presence of coils stimulates thrombotic mechanisms, contributing to the formation of a clot and leading to the cessation of blood flow to the chorioangioma, thereby treating the primary pathology.

Keywords

- ▶ giant chorioangioma
- ▶ microcoil embolization
- ▶ minimally invasive
- ▶ pregnancy outcome
- ▶ tumor devascularization

Introduction

Placental chorioangioma (CA) is the most common benign tumor of the placenta, seen in around 1% of placentas examined by microscopy. However, clinically significant “giant CAs,” which are > 4 cm, are less common, with an estimated incidence between 1:3,500 and 1:9,000 births (0.29–0.11%) with a female preponderance.¹ Studies have shown that giant CAs are associated with maternal, fetal, and neonatal complications, which include polyhydramnios, maternal preeclampsia with

mirror syndrome, preterm delivery, fetal growth restriction, fetal anemia, thrombocytopenia, fetal cardiac failure, hydrops, and fetal demise. Neonatal complications are microangiopathic hemolytic anemia, thrombocytopenia, and neonatal death.^{2–7} Early diagnosis, close prenatal surveillance, intrauterine intervention, and timely delivery would play a vital role in improving pregnancy outcomes.

The exact natural history and pathophysiology remain elusive, with various mechanisms proposed by different authors. The basic concept is that there is a preferential blood

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flow to the CA and away from the intervillous space, leading to diminished blood supply to the developing fetus (steal phenomenon) and thus a competitive substrate consumption by the tumor.⁸ Increased flow in the tumor leads to microangiopathic hemolysis and red cell sequestration, leading to severe anemia in the fetus. This leads to high output cardiac failure, hydrops, and fetal loss. Though spontaneous thrombosis and resolution of giant CAs resulting in a favorable pregnancy outcome have been reported in the literature,⁹ it is a rare event with most of the giant CAs causing adverse outcome in pregnancy leading to a 30 to 40% incidence of perinatal mortality.

The ideal and gold standard treatment remains a challenge as there is a paucity of data regarding the interventions available for managing large and clinically significant placental giant CAs. Amniodrainage, which is one of the conventional methods of treatment, is used to reduce amniotic fluid volume and thus reduces the intrauterine pressure in those cases associated with polyhydramnios. Cordocentesis and intrauterine blood transfusion of packed red cells were used in those cases associated with fetal anemia. However, these treatment methods provide only a symptomatic approach aiming to correct only the complications of CA and do not tackle the underlying pathology.

So, the primary treatment goal is the tumor's devascularization, which will address the underlying pathophysiology by disrupting the tumor's blood supply, thereby preventing the steal, substrate consumption, sequestration, and hemolysis. Various devascularization techniques have been tried and tested with varied outcomes; few have stood the test of time. Different methods used for the disruption of the tumor's blood supply are bipolar cautery, interstitial laser, radiofrequency ablation (RFA), fetoscopic laser photocoagulation, occlusion of feeder vessel using vascular clips under fetoscopic guidance, and embolization with liquid agents such as ethanol, cyanoacrylate, and embucrilate.¹⁰ Wide uptake of these procedures is limited due to their inherent risks and limitations. In this setting, ultrasound guided antenatal microcoil embolization of the feeder vessels could emerge as a novel, minimally invasive, potentially safe, and effective treatment option for symptomatic CAs.

Here, we report the successful management of a prenatally detected case of a large placental CA with fetal anemia and polyhydramnios using microcoil embolization followed by close surveillance and multidisciplinary care.

Case Report

A 34 year old second gravida with previous lower segment cesarean section was referred to us at 28 weeks' gestation in view of a placental CA for further evaluation and expert management. Her nuchal translucency (NT) scan at 13 weeks was normal, with the NT measuring 1.5 mm and no markers of aneuploidy. First trimester combined aneuploidy screening was not done and the targetted anomaly scan at 18 weeks showed a structurally normal fetus with optimal growth, liquor and dopplers.

At 28 weeks, an ultrasound done in our fetal medicine unit confirmed a hypoechoic well-circumscribed mass measuring

5.7 × 5.5 cm with a 6-mm feeder vessel originating very close to the cord insertion of an anterior placenta and seen coursing around the tumor mass before entering into it, the features which were consistent with a placental CA with associated polyhydramnios (single deepest pocket 13 cm). Color Doppler imaging revealed large vascular channels around and within the tumor. Middle cerebral artery (MCA) peak systolic velocity (PSV) was 1.7 MoM, suggestive of fetal anemia. There was no evidence of valvular regurgitation, cardiomegaly, or hydrops. The growth of the fetus was within normal limits and the umbilical artery and ductus venosus Doppler showed normal flow.

A multidisciplinary medical board was conducted involving the patient and her family, primary obstetrician, fetal medicine specialist, interventional radiologist, and neonatologist. She was explained the options of RFA versus microcoil embolization of the feeder vessel of placental CA along with amniodrainage and intrauterine blood transfusion. The risks of preterm premature rupture of membranes (PPROMs), abruption, preterm labor, cord complications, and intrauterine fetal demise if left untreated were also explained to the family. A 20% risk of recurrence of fetal anemia even after RFA/microcoil embolization was also discussed.

The ingenious nature of the planned intervention was also well explained to the patient and her family. The biggest challenge in front of us was that the feeder vessel was very close to the umbilical cord insertion site. So, under the aegis of the interventional radiologist, we decided for an ultrasound-guided microcoil embolization instead of RFA of the feeder vessel of the placental CA. Antenatal corticosteroid coverage was given before the procedure because of possible risks of fetal bradycardia, PPRoms, antepartum hemorrhage, and crash cesarean section in case of any adverse effects.

After informed consent, under monitored anesthesia care, and ultrasound guidance, the feeder vessel to the CA was targeted using an 18G Chiba needle. After confirming the intravascular position of the needle, an 8-mm Cook's MicroNester embolization coil (Cook Incorporated, United States) was deployed into the vessel. A 3-mm coil was also deployed to occlude a small branch of the feeder vessel. The size of the coil was decided based on the size of the feeder vessel. To prevent distal migration or embolization, it was decided that the first coil should be at least 2 mm oversized or 20 to 30% larger than the target vessel. The length of the coil depends on the length of the vessel, which was included in the embolization process. Post coil embolization, Doppler confirmed no vascularity within the tumor. The fetal heart rate remained stable throughout the procedure without evidence of any deterioration. The needle was withdrawn under ultrasound guidance and there was no evidence of extravasation of blood into the tumor or extraamniotic space. Amniodrainage was done immediately postprocedure and sample was sent to ascertain chromosomal normalcy as per the parents' request. Intrauterine blood transfusion was deferred as MCA PSV showed no evidence of severe fetal anemia. Postoperative ultrasound on day 1 showed a viable fetus, the coils in place without any migration, and no

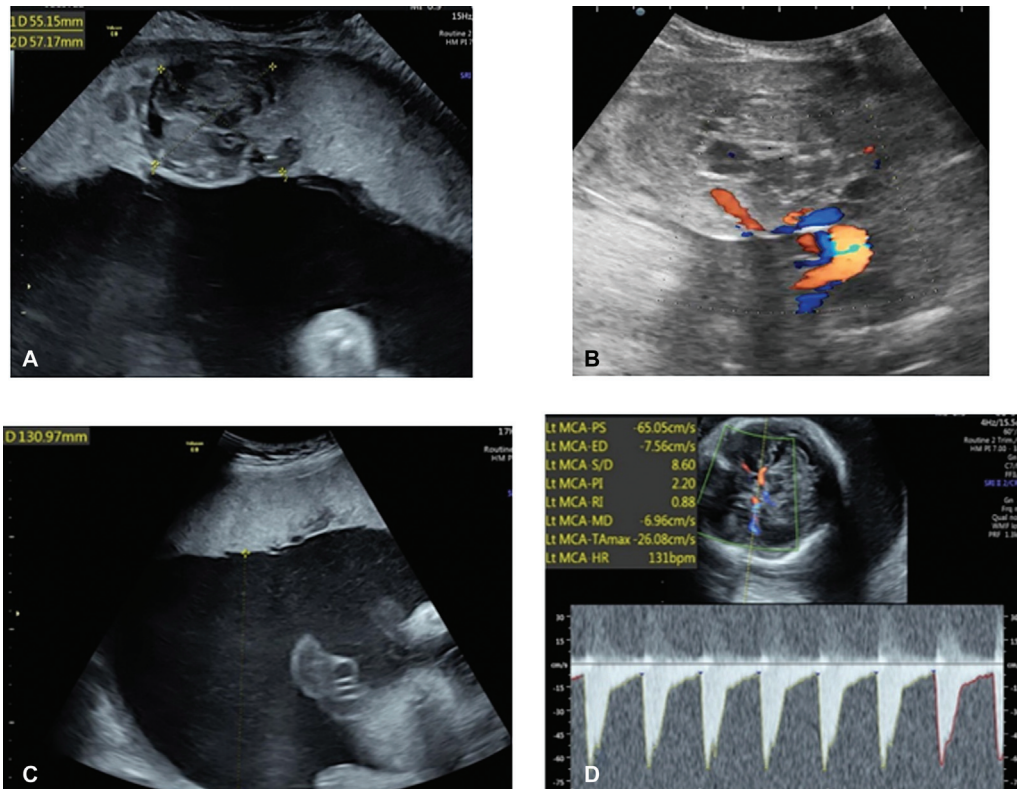


Fig. 1 Preembolization images. (A) Placenta chorangioma in gray scale. (B) Color Doppler showing feeder artery. (C) Polyhydramnios single deepest pocket (SDP) 13.9 cm. (D) Middle cerebral artery (MCA) peak systolic velocity (PSV) >1.5 MoMs → s/o fetal anemia.

evidence of flow within the tumor. Post procedure progesterone support was given.

We continued monitoring her pregnancy and her chromosomal microarray was reported to be a normal study. The amniotic fluid index (AFI) returned to normal (AFI 11.4), and fetal anemia was corrected with watchful expectancy alone. Baby's growth velocity continued to be normal and the Dopplers were optimal throughout the rest of her pregnancy. She delivered a live term healthy baby at 39 weeks weighing 3,650 g by cesarean section. Gross examination of the placenta revealed the CA with microcoils in situ and placental histopathology confirmed the same with lakes of blood vessels with angiomatous pattern, extensive necrosis, sclerosis, and hyalinization suggestive of placental CA with post

coil embolization status. The pre-embolization images of the CA are shown in ►Fig. 1A–D, microcoil embolization procedure images in ►Fig. 2A–C, the post microcoil embolization images in ►Fig. 3A and B, and ►Fig. 4A and B showing gross and histopathology images of the CA.

Discussion

The ideal management of placental CA still remains undefined. Conventional interventions like amniocentesis which aims at relieving the intrauterine excess pressure and cordocentesis with intrauterine blood transfusion which aims at correction of fetal anemia do not tackle the underlying pathophysiology. However, novel treatment procedures

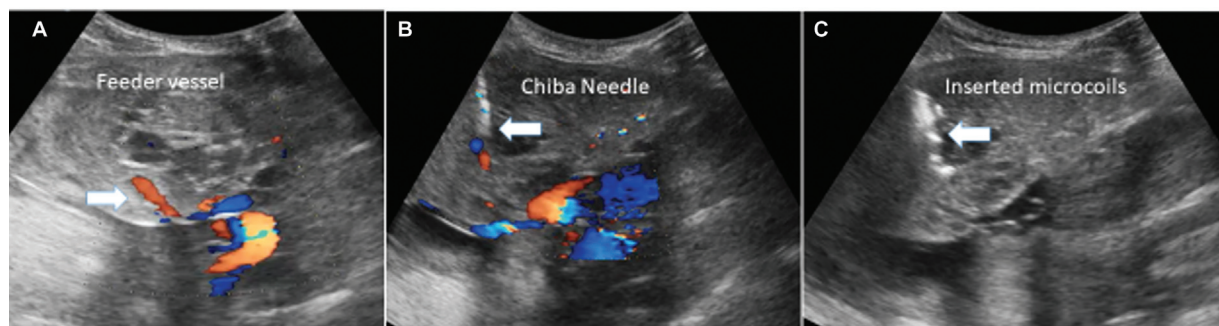


Fig. 2 Microcoil embolization images. (A) Shows the feeder's vessel. (B) Shows the introduction of the Chiba needle into the feeder's vessel. (C) Shows inserted microcoils in situ.

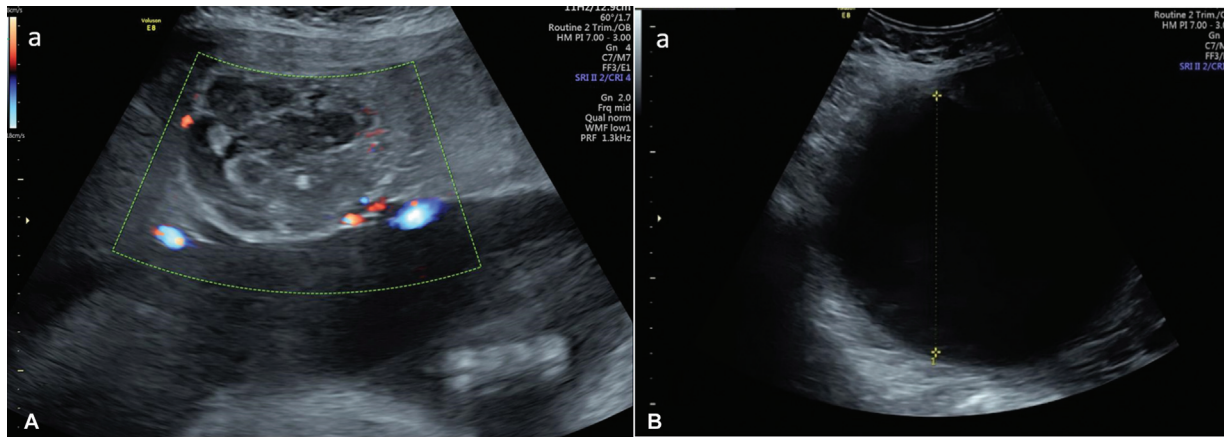


Fig. 3 Post microcoil embolization. (A) Color Doppler imaging shows no vascularity in the feeder vessel. (B) Reducing single deepest pocket (SDP) 8 cm.

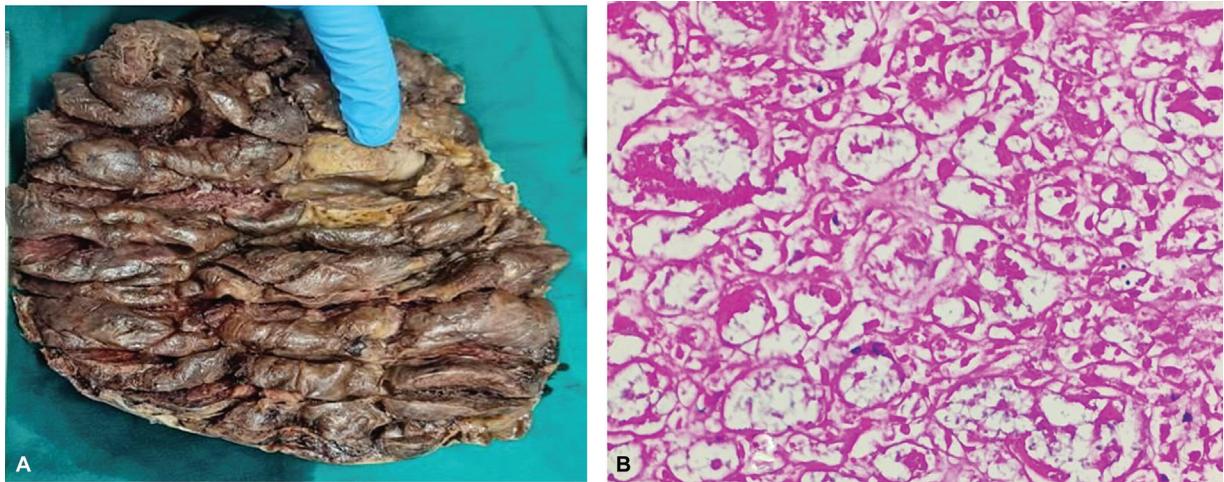


Fig. 4 Gross and histopathology images. (A) Gross picture of placenta with the chorangioma. (B) Histopathology image showing lakes of blood vessels with angiomatous pattern and extensive necrosis, suggestive of Chorioangioma with post coil embolization effect.

aim to address the cause and there have been various methods of tumor devascularization using liquid agents (ethanol, cyanoacrylate, and enbucrilate), different types of thermal energy such as bipolar cautery, laser, and RFA. Placing vascular clips under fetoscopic guidance have also been tried.¹⁰ Manikandan et al, in their review of various treatment modalities for placental CA, had concluded that no single intervention can be considered as the best fit for all cases of CAs.¹¹

The disadvantage of using liquid agents for feeder vessel occlusion is that placental CAs may shelter a few arteriovenous anastomoses, which allow passage of such embolic agents through the tumor to the fetus.¹² Voon et al reported a case of neonatal portal vein thrombosis after the use of the liquid embolic agent enbucrilate and advised vigilance while using this material.¹³ RFA and laser ablation procedures have the risk of accidental ablation of fetal cord vessels and heat transmission to nearby structures, causing unintentional side effects, fetal bleeding, exsanguination, and even fetal death, thus precluding their widespread use.

MicroNester embolization coils are made of platinum with spaced synthetic fibers, are available in a wide range of vessel sizes lengths from 3 to 20 cm and diameters from 2 to 20 mm, and are used for venous and arterial embolization in the peripheral vasculature. Though the evidence for using microcoils in obstetrics is limited, it is widely used for adequate thrombosis in various vascular pathologies. The mechanism of action is that the vessel stretch induced by the coil causes mechanical blockage of the vessel, while the fibers trap the circulating platelets, resulting in thrombus formation and cessation of blood flow in the feeder vessel.¹⁴

Very few case reports using microcoil embolization have been described in the literature. Lau et al in 2003, outlined the use of microcoils in the management of a giant CA (10 cm) with multiple small coil sizes ranging from 2 to 6 mm into the feeding vessel (size 7.4 mm) only to be repeated a week later using nine similar coils as complete occlusion was not achieved.¹⁴ The reason for repeat microcoil embolization was probably the inexact initial choice of smaller coil size than the size of the feeder vessel which

Table 1 Review of microembolization case reports

Author	Year	No. of cases	Size of the chorioangioma	Size of the feeder artery	Size of the coil used	Gestational age	Gestational age at delivery	Outcome
Lau et al	2003	1	10 cm	7.4 mm	2–6 mm	24 wk	29–30 wk	Neonatal death
Emery et al	2018	2	(1) 11.2 cm (2) 9.2 cm	9 mm 6 mm	6 mm 6 mm	26 wk 22 wk	26 wk 39 wk	Intrauterine fetal demise Live born
Present study Sabu et al	2023	1	5.7 cm	6 mm	8 mm	28 wk	39 wk	Live born

resulted in inadequate occlusion. Emery et al in 2018, described two cases of placental CA managed by microcoil embolization using two 6 × 2 mm Tornado microcoils for a 6-mm lumen primary feeding vessel and achieved rapid and complete occlusion,¹² which reiterates the importance of the right choice of the size of the microcoil. Out of these two cases where microcoil embolization was done, one of the pregnancies had a successful outcome and the cause of the intrauterine demise in the other fetus was attributed to the already sick and hydropic fetus where the procedure was not helpful. In our case, an 8-mm MicroNester embolization coil was deployed into the main feeder vessel which was 6 mm, and a 3-mm coil was introduced into a small branch of the feeder vessel ending in a successful and complete occlusion leading to an optimal perinatal outcome. Though the choice of the size of the coil is left to the discretion of the interventional radiologist/fetal medicine specialist, a coil size similar to the target vessel size or 2 mm bigger or 20 to 30% more than the target vessel would favor adequate occlusion of the feeder vessel^{15,16} as seen in our case and Emery et al. The details of the available microembolization case reports and the present study are presented in **Table 1**.

In this setting, ultrasound guided antenatal microcoil embolization of the feeder vessels could emerge as a minimally invasive, potentially safe, and effective novel treatment option for symptomatic CAs. This procedure can be performed entirely extra amniotic in anterior, lateral, or fundal placentae, thereby reducing the risks of amniorrhexis and its sequelae. The advantages of this procedure are (1) technical ease and feasibility as it is comparable to fetal blood sampling and intrauterine blood transfusion, (2) minimally invasive nature as only intravenous sedation/local anesthesia is needed and the approach being extra amniotic with an 18G needle, the risks of miscarriage/preterm birth can be reduced, (3) initiation of clot formation is at the desired site only with no migration through the tumor, and (4) lack of collateral damage to the adjacent structures which can occur with thermal energy.¹² However, the major limitation would be that the microcirculation supplied by the collaterals after occlusion of the feeder vessel might not be obliterated completely, especially if the size of the CA is large.¹⁴ This has been explained by Lau et al in their case report. The other disadvantages are the occlusion of nontarget vessels, coil migration, vessel perforation, infection, and allergic reactions, to name a few. However, as there are very few case reports of microcoil embolization around the world, the

exact incidence of these complications cannot be emphasized.

Conclusion

Large, symptomatic placental CAs if left untreated can lead to 30 to 40% perinatal mortality and there is a paucity of data regarding the optimal management. Literature review across the world reports very few cases using microcoil embolization. Our successful experience, the first of its kind reported in the country, opens the door to exploring the prospects of this procedure in anterior, lateral, and fundal placentae. Preoperative, intraoperative, and postoperative interdisciplinary collaborations are of prime importance in achieving successful outcomes. Intravascular access by interventional radiologist/fetal medicine specialist, precise selection of the size, number of coils, and location of placement play a crucial role in the successful management of such CAs. Microcoil embolization could emerge as a minimally invasive, potentially safe, and effective novel treatment option for symptomatic CAs. However, more prospective multicentric studies across the world are required to accurately establish the clinical safety and efficacy of microcoil embolization.

Conflict of Interest

None declared.

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