

A Large Placental Chorioangioma with Good Perinatal Outcome

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Chorioangioma is one of the most frequent benign neoplastic diseases of the placenta. Large chorioangioma is a rare primary placental tumor and usually measures greater than 5cm.

A twenty-six-year-old Filipina primigravida was seen at 29 weeks of gestation. At 37 weeks of gestation, she was noted to have a large 8×8 cm placental chorioangioma by ultrasound. She underwent elective cesarean section and delivered a live baby girl with good Apgar score. Histopathology of the placenta confirmed the diagnosis of 14×13×4 cm chorioangioma.

In this case, a large placental chorioangioma was found with good maternal and fetal outcome.

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Placental tumors are classified into trophoblastic and non-trophoblastic tumors. Chorioangiomas are the most prevalent non-trophoblastic type of tumors. These tumors are a common primary benign group^{1,2}. The incidence of chorioangiomas approximately ranges from 0.5% to 1%^{1,2,3,4,5}. Tumors that measure more than 5 cm are classified as large chorioangiomas. The occurrence varies in different studies between 1/2000-3500 births and 1/9000-50000 pregnancies^{3,5,6,7}. However, large chorioangiomas are rare. Most studies found a higher prevalence in primigravida, female sex fetus, twin pregnancy, elderly maternal age and other maternal morbidities, such as diabetes and hypertension^{4,8}.

The pathogenesis of this benign tumor is not well understood⁸. Hypoxia and genetics are factors cited in some studies^{9,10,11}. Histologically, these tumors are classified into angiomatous, degenerative and cellular. Angiomas are the most common form. Malformation, abnormal vascularization, arterio-venous shunts, proliferation and perfusion of the vascular system of the placenta lead to the prenatal complications⁸. Small chorioangiomas are mostly undiagnosed unless routine histopathological examination of placenta is performed. Small chorioangiomas are symptomless and mostly do not cause any maternal and fetal adverse events. On the other hand, larger chorioangiomas are often diagnosed during a routine antenatal ultrasound scan, mostly seen as a hypoechoic mass which contains anechoic cystic spaces in-between. The diagnosis accuracy could be confirmed by MRI imaging. Differential diagnosis includes partial molar pregnancy, retroplacental hematomas, submucous fibroid, and placental teratoma¹. Chorangiomas and chorangiomas are documented as a differential diagnoses because of their similar nature⁸.

These large tumors are prone to maternal complications and adverse fetal outcomes due to abnormal vascular shunts and vascular perfusions. Maternal complications include premature labor, polyhydramnios, abruptio placenta, preeclampsia, eclampsia and postpartum hemorrhage^{1,8,12}. Fetal complications include preterm birth, intrauterine growth restriction, fetal congestive cardiac failure, fetal hydrops, fetal anemia, fetal thrombocytopenia and intrauterine fetal death^{1,8,12,13}. These vascular placental tumors and their relationship with congenital malformations and chromosomal abnormalities were documented in few studies^{8,14,15}. Perinatal death of 30-40% was documented^{3,12}. Prognosis varies according to the size of the tumor, the degree of the vascular shunts and maternal and fetal complications^{6,12}.

The small size and asymptomatic tumors are usually managed conservatively with regular monitoring by ultrasound and assessment. Meanwhile, large tumors need frequent ultrasounds, Doppler studies for the assessment of vascular shunts, vascularity of the tumors, and middle cerebral artery, fetal cordocentesis and fetal echocardiogram to assess the fetal complications^{3,4}. Management could be amnio-reduction, intravascular transfusion, endoscopic laser coagulation, and embolization, interstitial laser therapy and injection of alcohol¹². The high rate of perinatal mortality justified the intrauterine interventions¹⁶. Most studies confirmed the need for close observation¹⁶. Recurrence of chorioangiomas is not well recognized^{8,17}.

The aim of this presentation is to report a case of large chorioangioma with its maternal and fetal outcome.

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THE CASE

A twenty-six-year-old Filipina primigravida presented at 29 weeks of gestation. The antenatal scan revealed a single active fetus with parameters corresponding to gestational age of 29 weeks of gestation. The estimated fetal weight was 1200 grams, which was an average for the gestational age. The placenta was reported to be not low-lying and no mass was mentioned. The patient was advised a detailed fetal anomaly scan, but she declined. At 37 weeks, ultrasound (US) revealed fetus with cephalic presentation. The estimated fetal weight was 2.5 kg. The amniotic fluid index was normal and no evidence of cardiomegaly, fetal ascites or hydrops. The placenta was fundal and it showed the rounded hypoechoic mass approximately 8 cm x 8 cm with anechoic cystic areas in-between near the cord insertion; these features are suggestive of a large chorioangioma, see figures 1,2,3. Umbilical arterial (UA) Doppler flow indices were normal, see figure 4.



Figure 1



Figure 2



Figure 3

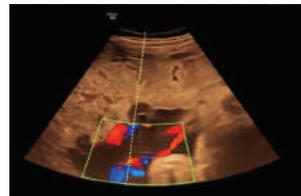


Figure 4

Figure 1: Ultrasound Image of Giant Chorioangioma

Figure 2: Ultrasound Image of Chorioangioma Near the Cord Insertion

Figure 3: Ultrasound Image of Chorioangioma Showing the Origin from the Placenta

Figure 4: Umbilical Artery Doppler Flow

Gestational age by US calculation was 35 weeks and 0 days. The sonographic biophysical score was 8/8. Placenta was anterior, showed a rounded hypoechoic mass with interspersed cystic spaces. The lesion was very vascular, measured 9x7cm and was seen in the upper end and reported as chorioangioma. The patient was admitted at 37 weeks and underwent cesarean section due to failed induced labor. The outcome was an alive baby girl with birthweight of 2260 grams and Apgar score of 9 at one and five minutes.

Intra-operatively, a complete separation of the placenta with large retroplacental clots and the liquor was noted to be bloodstained, see figure 5 and 6.

The postnatal period was uneventful. Postoperative hemoglobin was 9.2 grams. The baby was discharged in a stable condition after four days.

Histopathology report confirmed the diagnosis. The placenta measured 20x18x5cm and weighed 835grams. The umbilical cord showed three blood vessels. Microscopic examination

showed the placenta to have a benign vascular tumor composed of small capillary-sized blood vessels lined by plump endothelial cells with a slight increase in mitotic figures. The tumor involves approximately 60% of the tissue volume, see figure 7, 8, 9.

Postnatal follow-up after 6 weeks, the mother was doing well and breastfeeding. The baby was healthy weighing 4900 grams at 2 months of age.



Figure 5

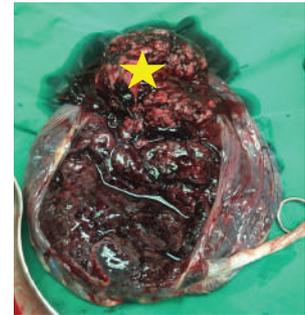


Figure 6

Figure 5: Gross Appearance of Placenta with the Vascular Mass Showing the Approximate Size

Figure 6: Gross Appearance of Placenta with the Globular Mass Showing the Retroplacental Clots

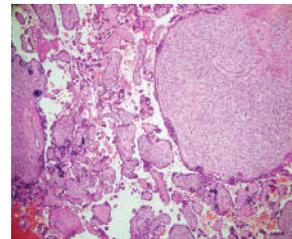


Figure 7

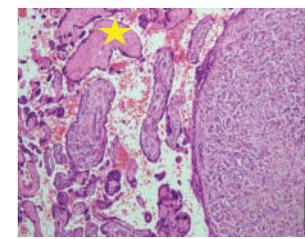


Figure 8

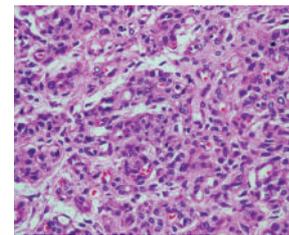


Figure 9

Figure 7: Proliferation of Capillary Sized Vessels Causing Expansion of Contiguous Affected Villi Along with Normal Villi

Figure 8: Proliferation of Capillary Sized Vessels Causing Expansion of Contiguous Affected Villi Along with Normal Villi

Figure 9: Proliferation of Capillary Sized Vessels Lined by Endothelial Cells

DISCUSSION

Although chorioangiomas are common benign tumors of the placenta, large chorioangiomas are rare^{3,5,6,7}. There are few reported cases of large-sized tumors measuring more than 12 cm. Generally, chorioangiomas are located near the umbilical cord insertion underneath the fetal surface¹⁸. Our case is unique given the large size of the tumor at 14x13x4cm. However, it was

typically located near the insertion of cord. Bisht et al reported a large case of 12x13x14 cm placental chorioangioma. Similarly, a larger size tumor of (20x18x17 cm) and its clinical implications were reported by Lau et al⁶. Presenting symptoms are frequently related to the size of the tumor and most of the small tumors are asymptomatic. A study showed that all the cases of small chorioangiomas were asymptomatic¹⁹. Common presentations of a large tumor are abdominal distension, abdominal pain, antepartum hemorrhage and preterm labor^{2,7,17}. Bashiri et al found a correlation between chorioangioma and the risk of preterm delivery²⁰. Our case was asymptomatic throughout her pregnancy even though the size of her tumor was considerably large. Her antenatal period was uneventful. She did not go into preterm labor. The fetus did not have any morbidities and showed no sign of adverse events or abnormalities.

It is presumed that this tumor starts to form around the 16-17th day of the evolution of the fetus²¹. The diagnosis of the tumor is usually by US after the second trimester^{7,16}. Elevated levels of maternal serum Alpha-Fetoprotein may help the diagnosis in early pregnancy²¹. The vascularity changes and the pulsatile flow of Colour Doppler in umbilical circulation and MRI are the other imaging modalities used to give the additional information of fetal circulation and to confirm the diagnosis. These imaging methods also help to diagnose the complications and differentiate this condition from the other similar conditions, such as placental teratoma, retroplacental clots and myomas. A report by Aaron et al confirmed the importance of color Doppler to differentiate chorioangiomas from other pathologies, such as hematomas¹.

Maternal and fetal complications of chorioangioma and its perinatal outcome were documented. A large chorioangioma causing fetal demise was reported²². A large placental chorioangioma with polyhydramnios was reported; the patient went into preterm labor and resulted in death from disseminated intravascular coagulation (DIC)². Bisht et al reported a case of chorioangioma and polyhydramnios without adverse events¹⁶. A similar case was diagnosed at 36 weeks of gestation with polyhydramnios, which was managed conservatively and timely intervention led to good outcome⁷.

The need to intervene during pregnancy depends mainly on the presence of complication which is directly related to the tumor size. Anyhow the aim of the intervention is ultimately to reduce the blood flow to the tumor and also to improve fetal hemoglobin^{6,12}. A retrospective study of 19 cases of giant chorioangioma revealed the association between the giant chorioangiomas and fetal adverse effects, such as fetal growth restriction, fetal anemia, non-immune hydrops, stillborn and polyhydramnios in the majority of cases³. The same study proved the timely prenatal intervention, such as amnio-drainage, in-utero blood transfusion and interstitial laser treatment. Another case documented the diagnosis of a large chorioangioma and its association with fetal complications of fetal growth restriction, oligohydramnios and fetal anemia⁶. A complicated case of chorioangiomas managed via laparoscopic-assisted laser ablation and embolization had a successful outcome of a term baby¹².

Most small and asymptomatic tumors are without adverse events and could be managed conservatively. On the other hand, large tumors with complications are in need of interventions. A case of chorioangioma with fetal hydrops and maternal complications was reported; the amnio drainage and in-utero transfusions did not improve the fetal and maternal

complications and ultimately led to preterm delivery and neonatal death¹³.

The size of the tumor is the most important criteria for the outcome and prognosis. A case of chorioangioma was diagnosed in the second trimester of the pregnancy with severe polyhydramnios and was managed by regular monitoring, amnio-reduction, and tocolytics achieved a good outcome without fetal abnormalities⁴. Wou et al concluded that not all chorioangioma cases necessarily lead to adverse outcomes¹⁹. Although the tumor in our patient was large in size, the absence of complications allowed conservative approach. With timely intervention, we could achieve good maternal and fetal outcome without adverse events except for the low-birth weight of the baby. Even though we found the placental separation and retroplacental clots intraoperatively, it is difficult to verify whether this bleeding was due to the induction of labor or the tumor itself; most likely placental separation happened during uterine manipulation at delivery. Analysis by many authors support the expectant management of asymptomatic tumor and documented their good outcome^{1,12}.

CONCLUSION

Our case is unique as it was a large chorioangioma; one among the few cases reported in the literature, and was managed conservatively. There were no maternal or fetal adverse effects. Therefore, our conservative management plan was justified. Induction of labor at 37 weeks was to prevent any adverse events to the mother or fetus and perinatal mortality.

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Competing Interest: None.

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Ethical Approval: The study design was approved by the Ethical Committee and Research Centre in Bahrain Defense Force Hospital.

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