Case Report

Placental chorangioma

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ABSTRACT

Placental chorangioma is a rare tumor with a frequency of 1%. It presents as a solitary nodule or less frequently as multiple nodules on the fetal surface of the placenta or within the placental parenchyma. Placental chorangiomas are considered clinically significant when >4 cm. Fetal complications associated with placental chorangiomas include polyhydromnios, nonimmune fetal hydrops, fetal heart failure, cardiomegaly, intrauterine growth restriction, fetal anemia, thrombocytopenia, and fetal demise. Maternal complications such as preeclampsia, preterm delivery, and maternal mirror syndrome are also associated with placental chorangiomas. The pathophysiology behind the complications is yet to be fully elucidated, but a prominent role for arteriovenous shunting and sequestration of red blood cells and platelets by the chorangioma has been postulated. The case presented was a 20-year-old primipara at 36 weeks of gestation with live fetus and an incidental finding of mixed echogenic mass lesion within the placenta measuring 59 mm × 52 mm. She was planned for elective cesarean section but went into spontaneous labor and had uneventful vaginal delivery.

Key words: Kano; live birth; placental chorangioma; Pregnancy.

Introduction

Placental chorangioma is a rare tumor of the placenta with a frequency of 1%.[1] It is thought to originate as a malformation of the primitive angioblastic tissue of the placenta.[2,3] It presents as a solitary nodule or less frequently as multiple nodules on the fetal surface of the placenta or within the placental parenchyma.[3] Placental chorangiomas are not uncommon among primiparas and twin pregnancies and their rate increases with maternal age.[3]

Placental chorangiomas are usually asymptomatic and merely discovered as incidental findings. However, they can present with fetal and maternal complications when they are large or multiple in number.[2] Placental chorangiomas vary in size and numbers. This variation has a direct relationship with both fetal and maternal complications. They are considered clinically significant when >4 cm,[4,5] while other workers[6,7] suggested a cutoff of ≥5 cm. However, large (≥4–5 cm) or multiple chorangiomas have been found to be associated with fetal, neonatal, and maternal complications.[8] Clinically evident, so-called "giant tumors" of the placenta are uncommon with an estimated incidence of 1:3500 and 1:9000 birth.[9]

Complications associated with placental chorangiomas include polyhydramnios, preeclampsia, preterm delivery, nonimmune fetal hydrops, fetal heart failure, cardiomegaly, intrauterine growth restriction, fetal anemia and thrombocytopenia, fetal demise, and maternal mirror syndrome.[7,10-19]

The pathophysiology behind the complications is yet to be fully elucidated, but a prominent role for arteriovenous

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shunting and sequestration of red blood cells and platelets by the chorangioma has been postulated.[8] Maternal mirror syndrome is defined as maternal edema due to fetal hydrops.[15] Its association with large placental chorangioma has been reported by several workers.[20-22] Other clinical features that can be found in patients with maternal mirror syndrome are also found in patients with severe preeclampsia which include elevated blood pressure, proteinuria, elevated uric acid and creatinine, headache, and visual disturbance.[8]

Case Report

She was a 20-year-old primipara who presented for routine antenatal follow-up visit with no complaints.

The pregnancy was spontaneously conceived and wanted. She booked at 25 weeks of gestation with no complaint. Her booking parameters were: weight 80 kg and blood pressure 120/70 mmHg. Her blood group was O Rhesus D+ and her genotype was AS. The Venereal Disease Research Laboratory (VDRL) and hepatitis B virus screening were nonreactive and Retro viral screening (RVS) was negative.

She had an obstetric ultrasound scan (USS) which showed a single live intrauterine fetus in longitudinal lie and breech presentation with good cardiac activity. The placenta was posterior on the body with homogeneous echo texture. The amniotic fluid volume was clear and adequate at estimated gestational age of 28 weeks. Expected date of delivery was January 14, 2017.

At 36 weeks of gestation, a repeat obstetric USS done, which revealed a singleton live intrauterine fetus in longitudinal lie and cephalic presentation with good cardiac activity. The placenta was postero-fundal. It, however, showed a fairly roundish mixed echogenic mass lesion on its fetal part measuring 59 mm × 52 mm in size toward the center of the placenta. No separation was seen. The amniotic fluid was clear and adequate [Figure 1].

The patient was informed about the condition, and a repeat ultrasonography was requested for confirmation. She was also counseled for elective cesarean section at 38 weeks of gestation. The repeat ultrasonography revealed the same roundish mass within the placenta measuring 59 mm × 54 mm. No separation was seen.

She presented with complaints of labor pains at 38 weeks of gestation. On examination, she was found to be afebrile, not pale, not dehydrated, anicteric, and not cyanosed. There was no demonstrable pedal edema and no peripheral lymphadenopathy. The abdomen was uniformly enlarged. There was no area of tenderness. The liver and spleen were not palpably enlarged. The kidneys were not ballotable. The symphysis-fundal height was 36 cm. The fetal lie was longitudinal in cephalic presentation at left occipito-posterior position. The fetal heart rate was 146 beats/min and regular. Pelvic examination revealed normal external genitalia; the cervix was anterior, soft, and fully effaced. The cervical os was 6 cm dilated with membranes bulging. A patograph was opened for recording of fetal and maternal vital signs and the progress of labor. A wide-bore cannula was inserted. She had her blood ready in case of any emergency. The theater and the anesthetist were informed. The fetal and maternal vital signs were normal.

Four hours after admission into the delivery suite, she had the urge to bear down. On review, the cervix was found to be fully dilated. She was encouraged to bear down with each uterine contraction. She subsequently delivered a live

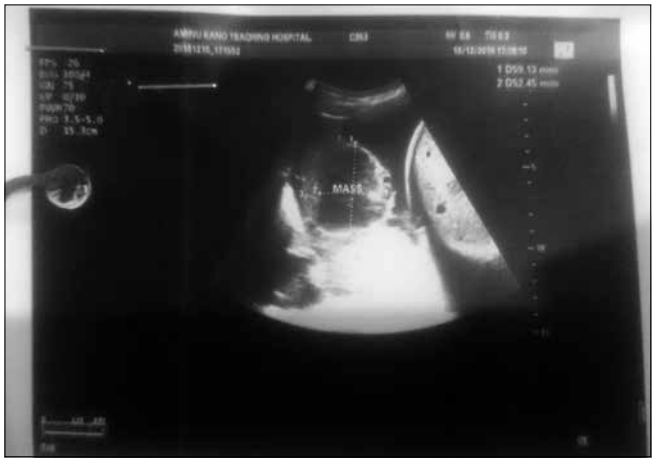

Figure 1: Complex mass of placental chorangioma protruding into the amniotic cavity

Figure 2: Placental chorangioma underneath the chorionic plate near the umbilical cord insertion

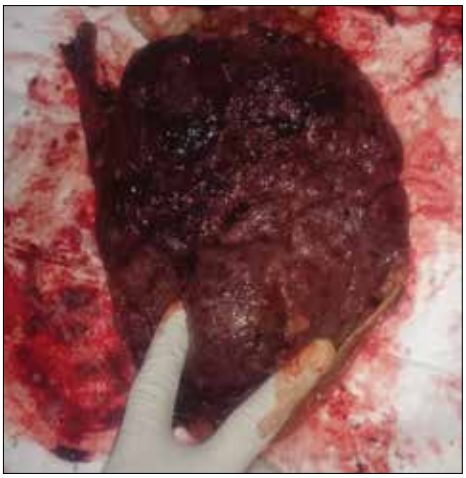

Figure 3: Maternal part of the placenta with chorangioma protruding (between the index and middle fingers)

female baby weighing 3.2 kg with Apgar scores of 8 and 9 at 1st and 5th min, respectively. The third stage of labor was managed actively. The placenta had a mass on its fetal surface which measured about 5 cm × 3 cm in diameter. It was firm in consistency [Figures 2 and 3]. The patient and relative were counseled on the need for histology. The placenta was sent for histology. Estimated blood loss after delivery was 150 ml. Histology result showed a well-circumscribed mass arising from stem villi. There was proliferation of capillary-sized vessels causing expansion of contiguous affected villi with associated areas of infarction. Other foci showed unremarkable villi.

Serum level of alpha-fetoprotein was normal.

Discussion

Clarke in 1798 first described placental chorangioma as hemangiomas.[23] Placental chorangioma is also referred to as placental angioma, chorionangioma, angiomyxoma, or vascular harmatoma of the placenta.[24] The tumor is frequently seen among Caucasians than African-American mothers and more often seen with multiple gestation.[25]

Diagnosis is usually by gray-scale sonography and by three-dimensional (3D) and 4D ultrasound during the antenatal period.[8] In this case, a gray-scale ultrasonography was used to make the diagnosis. Here, there was a fairly roundish mixed echogenic mass lesion on the fetal surface of the placenta. Placental chorangiomas are usually identified following USS as an hypoechoic, rounded mass, located near the chorionic plate, at times close to the umbilical cord insertion. It contains anechoic "cystic" lesion distinctly separate from the normal placental tissue. Degenerative processes within the mass may appear as heterogeneous areas with internal hemorrhage.[2] Chorangiomas may rarely appear as pedunculated.[2] Doppler velocimetry often shows a low-resistance pulsatile flow within the anechoic "cystic" areas representing enlarged vascular channels.[2] Large chorangiomas may undergo degenerative changes and present as areas of infarction with decreased echogenicity, decreased tumor volume, and decreased blood flow on color Doppler.[26] Color Doppler imaging (CDI), though available in our center, was unfortunately not requested for this patient.

Confirmation is by histology of the placental mass. Most chorangiomas are localized underneath the chorionic plate close to the umbilical cord insertion, protruding into the amniotic cavity.[8] This patient manifested with this clinical feature. Three histological types of placental chorangiomas are reported in the literature. These include the angiomatoid type which is characterized by numerous blood vessels and the degenerative type and the cellular type which have poor vascularization. Majority of the types tend to be sporadic.[2]

In low-resource settings, gray-scale ultrasonography is the only means of diagnosis. However, in developed nations, CDI is used to confirm the presence of vascular channels in the tumor, contiguous with the fetal circulation. These vascular channels can be used to differentiate chorangiomas from other placental lesions such as placental hematoma, degenerating fibroids, or partial mole.[19,27-30]

Usually, pregnancies with small chorangiomas progress with no complications, and as such do not require interventions.[8] Placental chorangiomas >5 cm or multiple in nature are associated with maternal, fetal, and neonatal complications, and as such, close surveillance is necessary to recognize early signs of serious complications and appropriate treatment offered.[8]

Management of symptomatic chorangiomas depends on the fetal status and the gestational age. Where pregnancy is toward term or fetal lung maturity is ascertain (gestational age >34 weeks), expedite delivery via a cesarean section is the preferred option. Alternative interventions for preterm pregnancies with placental chorangiomas include intrauterine blood transfusions of anemic fetus and amnio-drainage.[8] In this patient, pregnancy was uncomplicated, but we still planned elective cesarean section for her partly due to the size of the placental chorangioma coupled with the fear of fetal and maternal complications. Spontaneous onset of labor with good cervical changes resulted in vaginal birth of a live healthy baby before planned cesarean delivery.

Placental chorangiomas are infrequently associated with elevated maternal serum alpha-fetoprotein level. Khong and

George[31] reported that there was only one case of elevated maternal serum alpha-fetoprotein level among the 11 cases of placental chorangiomas they so far managed.

Both intrauterine blood transfusion and amnio-drainage do not affect and change the underlying pathophysiology of arteriovenous shunting within the chorangiomas. Targeted treatments to block arteriovenous shunting such as alcohol injection,[32] endoscopic laser coagulation, interstitial laser therapy,[33,34] and microcoil embolization[33] are being considered.

Conclusion

Placental chorangioma is a rare benign tumor. It can be diagnosed on routine 2D ultrasonography. Pregnancy with placental chorangioma may be carried normally, especially when it is solitary and <5 cm in dimension. There is a need to draw attention of the sonographer to be extra vigilant during USS in order to identify this rare tumor of the placenta.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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