Teenage Pregnancy with Chorioangioma: A Case Report with a Review on the Recent Trends on its Diagnosis and Management

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Abstract: Placental Chorioangiomas are rare placental tumors. They are often not diagnosed until delivery or till the onset of complications. A review of the literature reveals a wide spectrum of the presenting features and its associated complications. Usually, large tumors are associated with maternal and fetal complications. Recent advances in the diagnosis and management of these tumors have been worthwhile in cases associated with complications. Current advances in the management of large Chorioangiomas are noteworthy in cases of extreme prematurity as the timing of delivery is a challenge in such scenarios. However, the timing of delivery has to be individualized. Due to atypical presentation and unpredictable manifestations of Chorioangiomas, prompt diagnosis and timed follow up is mandatory in all cases. Though these tumors are usually associated with increased maternal age, we report a case of teenage pregnancy with Chorioangioma and its management.

Keywords: Choriongiomas, tumors, fetal medicine unit, complications, interventions.

INTRODUCTION

Chorioangiomas are rare placental tumors with unpredictable outcomes [1-3]. Large-sized tumors are associated with complications like polyhydramnios, placental abruption, pre-eclampsia, preterm labor, fetal anemia, thrombocytopenia, intrauterine growth retardation, fetal congestive cardiac failure, perinatal stroke and fetal demise [4-11]. Risk factors for chorioangioma include increased maternal age, first pregnancy, female fetus, high-altitude, essential hypertension and multifetal gestation [1, 2, 12]. Diagnosis is usually by ultrasound between 23-35 weeks of gestation and confirmed by histopathology [13-15]. Early diagnosis is preponderant as an increase in their size is an indication of potential complications. Though the management is usually conservative with 6-8 weekly serial ultrasound scans, however, some tumors grow aggressively and need to be reviewed weekly. Tumors larger than 4 cm in size may cause hydrops fetalis and its associated complications [3, 4, 17]. Management is individualized according to the gestational age and severity of complications [13, 14, 16]. Recent advances in the management of these cases are useful in cases of extreme prematurity as the timing of delivery remains a challenge for physicians in such scenarios. Though chorioangiomas are associated with increased maternal age, the following case report represents the successful management of teenage pregnancy with chorioangioma.

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CASE REPORT

18 years, young primigravida was booked in our antenatal clinic at 15 weeks of gestation. She did not have any significant medical and surgical history. She did not have family history of Chorioangioma. Booking laboratory investigations and viability scan was normal. She was diagnosed as a case of chronic urticaria at 18 weeks of gestation and was treated with antihistamines and oral steroids. At 24 weeks gestation, an anatomy scan revealed a hypoechoic mass in the placenta close to the cord insertion. For confirmation of the diagnosis, she was referred to the fetal medicine unit (FMU). She had another scan at FMU at 26 weeks of gestation which confirmed the diagnosis of chorioangioma. The tumor size was 3.45cm*3.2 cm with normal fetal growth and amniotic fluid index. She and her family were counseled in detail regarding the diagnosis and the potential risks associated and the need for close monitoring. She was offered a scan in FMU every 4-6 weekly. She was managed conservatively. Her glucose tolerance test at 26 weeks was within normal limits (Fasting- 4.0 mmol/L, 1st hour-6.0 mmol/L and 2nd hour-6.7 mmol/L). FMU scan was repeated after 6 weeks at 32 weeks 6 days of gestation and an insignificant increase in the tumor size was noted. No associated complications were noted.

The patient and her family were counseled in detail. She was on conservative management and another scan was scheduled after 6 weeks. At 37 weeks 4 days gestation she was admitted in labor. Labour was closely monitored and progressed well. She had an uneventful vaginal delivery. She delivered a healthy female baby, weight 3.392kg with APGAR score of 9/10. Active management of the third stage was performed. Placenta and membranes were examined

E-ISSN: 2309-4400/20

after delivery. Around 5 cm tumor with increased vascularity was noted in the placenta near cord insertion. Cord and cord vessels were normal on gross examination. The placenta was sent for histopathology. She was observed for post-partum hemorrhage for 24 hours. She had an uneventful post-natal period. She was discharged home in a clinically stable condition.

The histopathology result revealed an infarcted benign chorioangioma of the placenta. Infarction of chorioangioma impeded its growth and associated complications and thus the patient had an uneventful course during pregnancy.

DISCUSSION

Incidence

The incidence of chorioangioma is 1 % [1, 4]. Even though they are rare, but they are the commonest placental tumors [13, 18]. Large or multiple chorioangiomas are rarer with an incidence of 1:3500 to 1:16000 births [4]. Amer *et al.* has described Chorioangiomas as hamartomas [1]. It is suggested by Amala *et al*. that the massive proliferation of these tumors lead to arteriovenous shunts and may cause complications associated with them [4].

Risk Factors

Risk factors for chorioangioma include increased maternal age, first pregnancy, female fetus, high-altitude, essential hypertension, diabetes and multifetal gestation [4, 11, 25]. These tumors are usually associated with increased maternal age. Cases have been reported in women between 22-37 years of age with increased incidence with age, however, in the above case, it was associated with teen pregnancy. After reviewing the literature, it seems that this is the first case report associated with teenage pregnancy.

Pathophysiology

The exact pathogenesis of chorioangioma is not clear, however, it is suggested that the proliferation of primitive angioblastic tissue of the placenta may cause these malformations. Some authors have suggested the role of hypoxia and genetics in their pathology [4]. Other variants of cellular and degenerative types are also described by Amala *et al*. [4].

Diagnosis

These tumors are diagnosed by sonography; however, small tumors are often missed. In ultrasound scans, these tumors appear as well-circumscribed hypoechoic, circular areas near the insertion of the umbilical cord. Doppler study reveals low resistance pulsatile flow in the cystic areas [2, 13, 14, 19]. Serial scans may show accelerated growth and associated complications like polyhydramnios, hydrops, etc.]. In cases of large size tumors, other associated complications should also be evaluated during follow up scans. Because of the high risk for fetal heart failure associated with very vascular tumors, fetal echo should be offered and complete assessment of fetal color Doppler should be performed with the biophysical profile if indicated. Zanardini et at 2010, in their study, found the average gestation for diagnosis as 28 weeks + 4 days (ranging from 23 +weeks 2 days to 35 weeks + 1 day) [3]. Tumors markers, human chorionic gonadotrophin, and alpha-fetoprotein levels should be evaluated if there is any suspicion of trophoblastic neoplasia.

Differential Diagnosis

These tumors should be differentiated from molar pregnancies, placental site trophoblastic tumor and Choriocarcinoma as Doppler and ultrasound findings are similar to placental chorioangioma. Subamniotic hematoma, placental teratoma, atypical placental venous lake, etc. are few of the other differential diagnosis [4, 13, 14].

Complications

Tumors less than 4 cm diameter are usually benign [2, 5]. Large tumors and multiple tumors are rare but are associated with complications for both the mother and the fetus [5, 6, 16, 17]. However, very large tumors without any complications have also been reported [4]. Maternal complications like polyhydramnios may cause maternal distress, preterm labor, and placental abruption with higher incidence for the need for cesarean sections and postpartum hemorrhage [2, 3, 5, 26]. Fetal complications associated with chorioangiomas are fetal anemia, thrombocytopenia, intrauterine growth retardation, fetal congestive cardiac failure, and fetal demise [2, 3, 7, 9-11]. In the study by Zanardini *et al.* in a study of 19 cases, 7 patients (36.84 %) developed polyhydramnios. 31.58% (n=6) cases had growth-restricted fetus. 36.84 %(n=7) required prenatal intervention; fetoscopic laser treatment (n=1), amniodrainage (n=2) interstitial laser therapy (n=3) and elective preterm delivery (n=1), @32 weeks).

Interstitial laser therapy was given as early as 25 weeks to prevent fetal demise. The average gestational

age at delivery was 35weeks (33 weeks + 0 to 38 + 5weeks). One fetus had stillbirth [3]. Ghidini *et al.* in a case report discussed the association of chorioangioma with perinatal stroke on the third day of birth [8]. Several other studies have shown an association with perinatal cerebral arterial infarction [7, 9, 10].

Management

Chorioangioma usually has a benign course and is treated with expectant management [6]. Small tumors (less than 4 cm are usually monitored every 6-8 weeks with an ultrasound scan, whereas large highly vascular tumors require serial ultrasound examination with Doppler flow studies every 1-2 weeks. Treatment of complications is usually case-specific. Amnioreduction for polyhydramnios reduces maternal discomfort and minimizes the risk of preterm labor, rupture of membranes, placental abruptions and post-partum hemorrhage. Therapeutic amniocentesis and maternal indomethacin therapy are the other alternatives. Many recent studies have advocated the improvement in neonatal outcomes by ultrasound-guided or endoscopic laser devascularisation in the fetus with hydrops or early features of congestive cardiac failure. Jones *et al.* have recommended the injection of thrombogenic materials and microcoil embolization [24]. Other suggested measures are intratumoral alcohol ablation and laser surgery in thrombosing blood flow to a large placental chorioangioma [20]. Ercan *et al.* applied a combined approach with intratumoral alcohol injection, cordocentesis, intrauterine transfusion, and amnioreduction in a large placental chorioangioma [20]. Hamouda *et al.* suggested in utero embolization for placental chorioangioma and neonatal multifocal hemangiomatosis [21]. Radiofrequency ablation has been tried successfully by some authors [22, 23].

Timing of Delivery

For small tumors without any fetal or maternal complications, there is no need to induce labor or expedite delivery unless indicated for any other maternal or fetal reason. Some cases of chorioangiomas with without polyhydramnios are complicated with preterm birth [26]. The timing of delivery in these cases depends on the severity of complications and the gestational age of the fetus. In the second trimester or cases of extreme prematurity, delivery is not considered due to pre-maturity. In these cases, the offered management should be case-specific. In cases of polyhydramnios amnio-reduction [21, 25] and in case of severe fetal anemia intrauterine transfusions [21] should be offered at a feto-maternity specialized unit. In patients with progressively increasing/large tumors with complications, the available options are devascularisation of tumor, embolization of tumor blood vessels, laser coagulation or ablation with radiofrequency [20-22, 24, 25]. Chemosclerosis is also an option, where other methods are not available [27]. Post-treatment follow up scans should be monitored to assess the response and to address ongoing complications. In the third trimester delivery may be timed depending on the severity of the complications. Antenatal fetal surveillance is mandatory where invasive methods are offered. Antenatal steroids should be offered in preterm cases with elective delivery.

CONCLUSION

Chorioangiomas though uncommon but are associated with fatal maternal and fetal complications. Close monitoring by ultrasound and timely management of complications is the key to reduce neonatal and maternal morbidity and mortality. As they have a wide spectrum of etiology and unpredictable course of progression, these cases should be reported. Future literature reviews and systemic reviews would help understand the progression of these tumors.

CONFLICT OF INTEREST

No conflict of interest was declared by the authors.

FINANCIAL DISCLOSURE

None.

ACKNOWLEDGEMENTS

None.

REFERENCES

[1] Amer HZ, Heller DS. Chorangioma and related vascular lesions of the placenta — a review. Fetal and Pediatric Pathology 2010; 29(4): 199-206. https://doi.org/10.3109/15513815.2010.487009

[2] Al Wattar B, Hillman S, Marton T, Foster K, Kilby M. Placenta chorioangioma: a rare case and systematic review of literature. The Journal of Maternal-Fetal & Neonatal Medicine 2013; 27(10): 1055-1063. https://doi.org/10.3109/14767058.2013.847424

[3] Zanardini C, Papageorghiou A, Bhide A, Thilaganathan B. Giant placental chorioangioma: natural history and pregnancy outcome. Ultrasound in Obstetrics and Gynecology 2010; 35(3): 332-336. https://doi.org/10.1002/uog.7451

[4] Amala S, Veena N, Abdulla D, Nawal D. A Large Placental Chorioangioma with Good Perinatal Outcome. Bahrain Med Bull 2019; 41(2): 115-118.

[5] Kirkpatrick AD, Podberesky DJ, Gray AE, *et-al*. Best Cases from the AFIP: Placental Chorioangioma. Radiographics 2007; 27(4): 1187-90. https://doi.org/10.1148/rg.274065207

[6] Kodandapani S, Shreshta A, Ramkumar V, Rao L. Chorioangioma of Placenta: A Rare Placental Cause for Adverse Fetal Outcome. Case Reports in Obstetrics and Gynecology 2012; 2012: 1-3. https://doi.org/10.1155/2012/913878

[7] Mineyko A, Kirton A. The Black Box of Perinatal Ischemic Stroke Pathogenesis. Journal of Child Neurology 2011; 26(9): 1154-1162. https://doi.org/10.1177/0883073811408312

[8] Ghidini A, Locatelli A. Diffuse placental chorioangiomatosis causing multiple fetal cerebral embolism: a case report. J Reprod Med 2006; 51: 321-4.

[9] Das S, Ankola P, Chiechi M, Sandhu J. Perinatal Cerebral Arterial Infarction Associated with a Placental Chorioangioma. American Journal of Perinatology 2008; 25(6): 381-383. https://doi.org/10.1055/s-2008-1078755

[10] Lehman L, Rivkin M. Perinatal Arterial Ischemic Stroke: Presentation, Risk Factors, Evaluation, and Outcome. Pediatric Neurology 2014; 51(6): 760-768. https://doi.org/10.1016/j.pediatrneurol.2014.07.031

[11] Imdad A, Sheikh L, Malik A. A large chorioangiomas causing intrauterine foetal demise. J Pak Med Assoc 2009; 59: 580-1.

[12] Reshetnikova O, Burton G, Milovanov A, Fokin E. Increased incidence of placental chorioangioma in high-altitude pregnancies: Hypobaric hypoxia as a possible etiologic factor. American Journal of Obstetrics and Gynecology 1996; 174(2): 557-561. https://doi.org/10.1016/S0002-9378(96)70427-1

[13] Taori K, Patil P, Attarde V, Singh A, Rangankar V. Chorioangioma of placenta: Sonographic features. Journal of Clinical Ultrasound 2008; 36(2): 113-115. https://doi.org/10.1002/jcu.20366

[14] Lurain J. Gestational trophoblastic disease I: epidemiology, pathology, clinical presentation and diagnosis of gestational trophoblastic disease, and management of hydatidiform mole. American Journal of Obstetrics and Gynecology 2010; 203(6): 531-539. https://doi.org/10.1016/j.ajog.2010.06.073

[15] Gimenez A, Kopkin R, Chang D, Belfort M, Reece E. Advances in Fetal Surgery: Current and Future Relevance in Plastic Surgery. Seminars in Plastic Surgery 2019; 33(03): 204-212. https://doi.org/10.1055/s-0039-1693431

[16] Wu Z, Hu W. Clinical analysis of 26 patients with histologically proven placental chorioangiomas. European Journal of Obstetrics & Gynecology and Reproductive Biology 2016; 199: 156-163. https://doi.org/10.1016/j.ejogrb.2015.12.009

[17] Dong T, Sher D, Luo Q. Pregnancy complications and adverse outcomes in placental chorioangioma: a retrospective cohort analysis. The Journal of Maternal-Fetal & Neonatal Medicine 2018; 1-5. https://doi.org/10.1080/14767058.2018.1548598

[18] Quintero R, Reich H, Romero R, Johnson M, Gonçalves L, Evans M. In utero endoscopic devascularization of a large chorioangioma. Ultrasound in Obstetrics and Gynecology 1996; 8(1): 48-52. https://doi.org/10.1046/j.1469-0705.1996.08010048.x

[19] Zalel Y, Weisz B, Gamzu R, Schiff E, Shalmon B, Achiron R. Chorioangiomas of the placenta: sonographic and Doppler flow characteristics. J Ultrasound Med 2002; 21(8): 909-13. https://doi.org/10.7863/jum.2002.21.8.909

[20] Ercan C, Coksuer H, Karasahin K, Alanbay I, Baser I. Combined Approach in a Large Placental Chorioangioma Case with Intratumoral Alcohol Injection, Cordocentesis, IU Transfusion, and Amnioreduction. Fetal and Pediatric Pathology 2012; 31(6): 374-378. https://doi.org/10.3109/15513815.2012.659402

[21] Hamouda S, Soussan J, Haumonté J, Bretelle F. In utero embolization for placental chorioangioma and neonatal multifocal hemangiomatosis. Journal of Gynecology Obstetrics and Human Reproduction 2019; 48(8): 689-694. https://doi.org/10.1016/j.jogoh.2019.05.011

[22] Phithakwatchara N, Makarasen P, Nawapun K, Hanamornroongruang S, Viboonchart S, Wataganara T. Time and temperature dependence of radiofrequency ablation in the human placenta. Prenatal Diagnosis 2018; 38(7): 504-510. https://doi.org/10.1002/pd.5276

[23] Hun K, Nassar P, Chen T, Sardesai S, Chmait R. Giant Chorioangioma TreatedIn Uterovia Laser of Feeding Vessels with Subsequent Development of Multifocal Infantile Hemangiomas. Fetal and Pediatric Pathology 2014; 34(1): 1-8. https://doi.org/10.3109/15513815.2014.925018

[24] Jones K, Tierney K, Grubbs B, Pruetz J, Detterich J, Chmait R. Fetoscopic Laser Photocoagulation of Feeding Vessels to a Large Placental Chorioangioma following Fetal Deterioration after Amnioreduction. Fetal Diagnosis and Therapy 2012; 31(3): 191-195. https://doi.org/10.1159/000331944

[25] Kirstie M. Franceschina, Sonographic Evaluation of a Large Placental Chorangioma. Journal of Diagnostic Medical Sonography 2017; 33(3): 246-250. https://doi.org/10.1177/8756479317691357

[26] Bashiri A, Furman B, Erez O, Wiznitzer A, Holcberg G, Mazor M. Twelve cases of placental chorioangioma, Pregnancy outcome and clinical significance. Arch Gynecol Obstet 2002; 266: 53-55. https://doi.org/10.1007/PL00007501

[27] Wanapirak C, Tongsong T, Sirichotiyakul S, Chan-Prapaph P. Alcoholization: the choice of intrauterine treatment for chorioangioma. Journal of Obstetrics and Gynaecology Research 2002; 28(2): 71-75. https://doi.org/10.1046/j.1341-8076.2002.00016.x

Received on 28-01-2020 Accepted on 15-02-2020 Published on 28-02-2020

DOI: https://doi.org/10.31907/2309-4400.2020.08.02