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Giant Chorioangioma with Oligohydramnion: A Case Report

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ABSTRACT

Chorioangioma or chorangioma is the most common non-trophoblastic benign and angiomatous vascular tumor of the placenta with a prevalence rate of 0.6-1%. It was first described by Clark in 1798. The smaller types of less than 4-5cm occurring in less than 1:10,000 pregnancies tend to be asymptomatic and clinically not significant while larger chorioangiomas found in over 1:16,000 births could manifest with adverse maternal, fetal and neonatal outcomes. The tumor related manifestations in this case report encompass a multitude of features that displayed a placental mass of 8.5 x 8.6cm, oligohydramnion, intrauterine growth restriction, poor biophysical profile and a preterm breech that culminated in an emergency cesarean delivery. The intent of the article is to create awareness of the possible pathology among practitioners, raise a high index of vigilance and establish early diagnosis, prognosticate and enhance the concept of birth preparedness and complication readiness.

Keywords

Chorioangioma, Oligohydramnion, IUGR, Preterm, Breech presentation, Cesarean delivery, Addis Ababa, Ethiopia.

Abbreviations

BPP: Biophysical Profile, CD31: Cluster of Differentiation 31, CD34: Cluster of Differentiation 34, CDI: Color Doppler Imaging, IUFD: Intra Uterine Fetal Death, D (2,3,4): Ultrasound imaging as flat pictures , baby's features, live motions, EFW: Estimated Fetal Weight, ELLP: Elevated Liver Enzymes and Low platelet count, ENND: Early Neonatal Death, HBsAg: Hepatitis B Surface Antigen, HCV Ab: Hepatitis C Virus Antibody, HELLP : Hemolytic anemia Elevated Liver enzymes and Low Platelet count, HIV: Human Immunodefficiency Virus, IUGR: Intrauterine Growth Restriction, PIHCT: Provider Initiated HIV Counseling and Testing, VDRL: Venereal Disease Research Laboratory.

Introduction

The human placenta is a 15-20cm long, discoidal in shape and hemochorial fetal structure with 2-2.6 cm thickness at its center and peripheral thickness of 1.5 cm. The amnion is closely related to the fetal surface while the chorion is apposed to the maternal side. Placenta weighs about 500g corresponding to 1/5 or 1/6 of fetal weight. The surface area of the placenta is 14 m^2 and is 10 times greater than the body surface area. The blood circulation in the intervillous space amounts to 400-500 ml/min while that of the uterus is 500-700 ml/min. Under normal conditions, there is no mixing of fetal and maternal blood. The placenta is attached to the basalis layer, which forms the chronic plate establishing the feto-maternal unit [1-4].

The placenta produces enzymes like thromboplastin-thromocinase and oxytocinase, proteohormones (human chorionic gonadotropin, human placental lactogen, human chorionic somatomammotropin, and human chorionic thyrotropin) and steroid hormones (estrogen and progesterone). The parabiotic unity deters and prevents undue immunological reaction and rejection of the fetal allograft [1,2,4].

The placenta functions as a respiratory (supplying oxygen and removing carbon dioxide) and excretory (removing or filtering out harmful wastes) organs. As a fetal organ, it assumes the sole inherent metabolic exchange function of the liver supplying glucose and other nutrients; and moreover producing and maintaining hormones in the equilibrium that are beneficial to

the mother and fetus. It passes immunologic factors from the mother to the fetus; and along with the amniotic fluid creates a hydrostatic environment protecting the fetus. Hence, it ascertains the dictum of healthy placenta-healthy fetus relationship in lieu of the unfavorable impact of chronic placental insufficiency and hypoxia [1,4].

The non-trophoblastic tumors of the placenta comprise of chorioangioma (chorangioma), chorangiosis, hemangioma, teratomas, hamartomas, hematomas, leiomyoma and hepatocellular adenoma. The subtype and which is the commonest is chorioangioma. It was first described by Clark in 1798 and accounts for an overall prevalence of 0.6 - 1% with an incidence of 1:16,000-90,000 births. The smaller ones with an incidence of 1:500-10,000 are characterized as asymptomatic and not significant; and they assume clinical denotation only when they measure more than 4 - 5cm in size [5-10].

Chorioangiomas, the focal point in this presentation, are basically well demarcated structures found on the fetal surface or within the villous parenchyma. They characteristically protrude towards the amniotic cavity and most often are located at about the umbilical cord insertion. They are covered by a single or double layer chorionic epithelium. Chorioangiomas usually exist as a solitary nodule but yet expressed with occasional multiplicity unlike the larger ones which are quite rare [6].

The pathophysiological arteriovenous shunting and sequestration of red blood cells and platelets by the giant chorioangiomas may result in poly or oligo-hydramnion, non-immune hydrops fetalis, hemolytic fetal anaemia, thrombocytopenia, cardiomegaly, heart failure, intrauterine growth restriction, intrauterine fetal death and could as well be associated with early neonatal loss. The maternal related complications include pre-eclampsia, preterm delivery and maternal mirror syndrome (Ballantyne syndrome) with placentomegaly [6,11,12].

The anatomical, physiological and obstetrical challenges are briefly reviewed and the chorioangioma-related pathophysiological alterations and derangements on the placental functions as could be expressed in maternal, fetal and neonatal clinical presentations are highlighted. Hence, the above elaborated narrative on various aspects of human placenta in addition to giant chorioangioma/ chorangioma is relevant and consistent with the clinical presentation of the case in focus. It is of the first ever report within the Ethiopian context.

Case Description

We hereby present a 28 years old primigravida who has had three antenatal follow ups at the ET hospital, which is privately owned. She came one week prior to her appointment date at gestational age of 36 weeks and 3 days from the last normal menstrual period with a complaint of decreased fetal movement and absent fetal kick in the last 48 hours. She was normotensive with a body weight of 61kg, height of 158cm, and a BMI of 24.4Kg/m^2. She is economically well of and capable to afford treatment in a private setup.

The antenatal workup for routine fasting blood sugar, uric acid, complete blood count including blood grouping and Rh determination, urine analysis and serology for HBsAg, HCV antibody, PIHCT (provider-initiated HIV counseling and testing) and VDRL were all performed, and found to be non-revealing. In particular and of interest, there was no Rh setup. Based on the composite clinical presentation of the patient, she was then further investigated for Hemolytic anemia, Elevated Liver enzymes and Low Platelet counts in association with possible development of pre-eclampsia constituting HELLP and ELLP (without the hemolytic component) syndromes, respectively.

Ultrasound examination provided a fetus with a longitudinal lie and breech presentation with no obvious gross congenital malformation on further scanning for anatomical anomaly. The fetal biometry yielded a gestational age of 35 weeks and 4 days with HC/AC ratio of 1.06 and an estimated fetal weight of 2.3Kg. It was registered that the gross fetal movement, fetal tone and fetal breathing movements were all nil in 30min, respectively and the amniotic fluid pool was less than 5cm and interpreted as inadequate and labeled “severe oligohydramnion”. Hence, the summation of the components of the determined biophysical profile (BPP) of 0/8, short of contraction and non-stress tests was declared unfavorable and dictated the intervention.

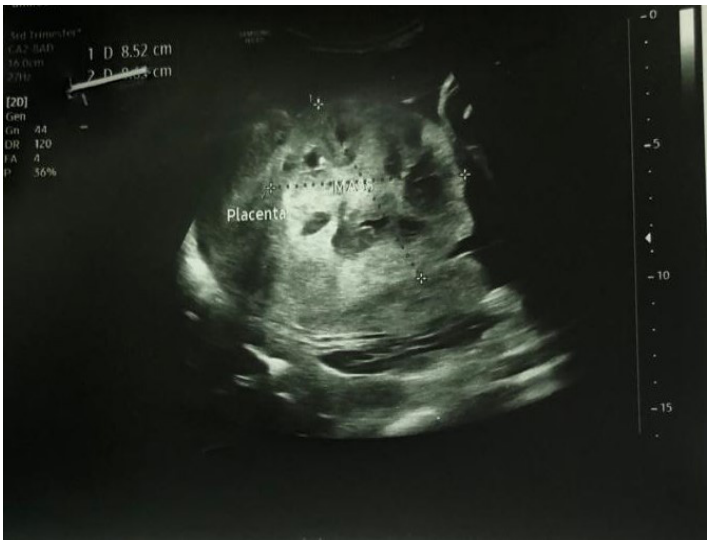
The placenta was located on the posterofundal aspect of the uterus with no low-lying component. The record documented an incidental finding of a well-defined predominately hyperechoic, hypervascular mass at the fetal side of the placenta measuring a placental mass of 8.5 x 8.6 cm in size as a probable diagnosis of a chorioangioma.

She underwent cesarean delivery for breech presentation, IUGR, oligohydramnion and extremely unfavorable biophysical profile (BPP). The outcome was a female neonate with Apgar score of 4/10, 6/10 and 7/10 in the first, fifth and tenth minutes, respectively and a birth weight of 2120gm.

On histopathological examination, the placenta measured about 14 x 14 x 6 cm while that of the placental subchorionic mass measured 9 x 8 x 5 cm with a weight of 1.4 kg. The umbilical cord measured 42cm and had two arteries and a vein with normal Wharton’s jelly; and further demonstrated no true or false knots. The histological report reaffirmed the diagnosis of chorangioma.

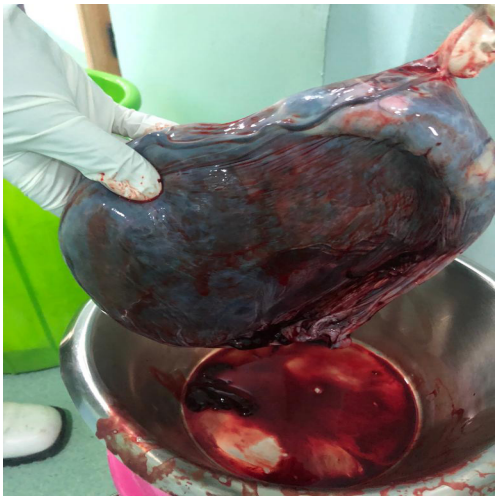
They both fared well and discharged on the third postoperative day in good health. Their subsequent postnatal follow up on the first and sixth weeks ensured smooth course with no complications and abnormalities.

The Ultrasound Picture of the Hyperechoic, Hypervascular Placental Mass



Ultrasonographic fetal anatomical scan for any anomaly was unremarkable but concluded the report as a third trimester pregnancy of 35w 4d, intrauterine growth restriction (IUGR), oligohydramnion, preterm breech presentation, fetal weight of 2.3kg with a poor biophysical profile (BPP) of 0/8, short of contraction and non-stress tests.

The Gross Appearance of the Placental Tumor



The record documented an incidental finding of a well-defined predominately hyperechoic, hypervascular mass at the fetal side of the placenta measuring 8.5 x 8.6 cm in size as a probable diagnosis of a chorangioma.

Pathology Report

Clinical history and findings

- Cesarean section done for poor BPP and preterm breech
- 8.5 x 8.6 cm placental mass on antenatal ultrasound
- Postpartum confirmation the placental mass

Gross findings

- Placenta: 14 x 14 x 6 cm, with a 42-cm umbilical cord
- Cross section: 9 x 8 x 5 cm dark-brown placental mass
- Normal-looking membranes and cord

Microscopy

Section from the placental mass shows expansion of the villous structures with proliferation of capillary-sized vessels with little intervening stroma. This is consistent with **chorangioma.**

Discussion

Chorioangioma or also referred to as chorangioma is a rare and commonest benign tumor of the human placental non-trophoblastic tumors. It was first described by Clark in 1798 but it was Beneke who for the first time designated the tumor as chorioangioma in 1900. The maternal and fetal clinical manifestations in our case include a large placental mass, oligohydramnion, IUGR, low birth weight, poor biophysical profile and breech presentation culminating in an emergency cesarean delivery. Chorioangioma in association with oligohydramnion has been reported as early as 1953 though polyhydramnion seems to be the rule, which in turn has to be scrutinized for placentomegaly, hydrops fetalis, multiple pregnancy and macrosomia with diabetes mellitus. Although the immediate outcomes and conditions upon discharge of both were free of complications, the article shall further revisit aspects of the antenatal, maternal, fetal and neonatal outcomes vis-à-vis the hallmark of the pathophysiology of larger chorioangiomas [13-15].

The risk factors enumerated upon, though not absolute, include age of 30 or more years linearly, primigravidity, multiple pregnancy, history of impaired glycemic profile, pre-eclampsia, female sex of the fetus in utero and preterm delivery. The chorioangiomas are mostly observed in the third and less frequently in the second trimester of pregnancy. The speculated and incriminated pathophysiological mechanisms for the genesis of chorioangioma in addition to the aforementioned predisposing elements that are worth mentioning are genetic factors, higher altitudinal hypoxic effects and hypoperfusion. We believe that some of these factors might have been in the interplay in our patient as Addis Ababa lies at an altitude of 2,355 meters above sea level [16,17].

Taking the clinical scenario in perspective, history and physical examination play an important role in keeping with the differential diagnosis when oligohydramnion occurs in conjunction with IUGR, a malpresentation, fetal and medical complications. A 2D, 3D, 4D and Color Doppler Imaging (CDI) are very helpful auxiliary diagnostic instruments as chorioangiomas possess the typical inherent capillarious structures. Histological studies definitely establish the final diagnosis. Further enthusiastic and sophisticated immunological and histochemical tests, where available, could be aspired for. Hence none of the following tests like CD31, CD34, factor VIII and GLUTI along with focal staining with cytokeratin that could be originating from the blood vessels was studied [10,18,19]. We are hopeful that one day, out of sheer necessity they shall be made available at least in the teaching and private institutions.

Expectant management with frequent antenatal visits, induction of lung maturity with corticosteroid administration, fetal kick chart monitoring, timely hospital admission and expedite delivery is intentionally well founded provided that one ensures the effect of chronic placental insufficiency and chronic hypoxia would not cause fetal comprise by worsening the IUGR, causing IUFD and/or contribute to ENND. There was no place for therapeutic amnion reduction/drainage as our patient had oligohydramnion instead of polyhydramnion. The up-to-date and ultramodern invasive options in the presence of the fetus in utero with fetoscopic laser coagulation/ablation, endoscopic surgical devascularization and chemosclerosis/alcohol injection could be attempted where the technology is available, the manpower exists, financial resources permit and follow ups are guaranteed. In a high parity population with low resource setting, the introduction of such advanced technology may be considered a luxury but should be introduced as an absolute necessity and made the services attainable [17,20,21].

Placental chorioangioma represents itself as a solitary or multiple, small or large nodules on the fetal surface or within the placental parenchyma. The smaller chorioangiomas are invariably asymptomatic and are often detected incidentally during placental examination. Large chorioangiomas may undergo degenerative secondary changes resulting in necrosis, infection, infarction, hyalinization, calcifications, fatty accumulation and myxomatous changes. In the presence of such a wealth of information on placental pathology, we must capitalize on the evaluation of the placenta especially in the antenatal period as it has greater influence in the subsequent management of the case [22-24]. All such cases need to be prospectively and thoroughly collected, documented, reported domestically, and internationaly.

The relationship of a healthy placenta with a healthy fetus not only determines that of the fetus but may affect the overall fetal and maternal morbidity and mortality. Hence, it is overwhelmingly evident that the authors highly advocate and recommend mandatory immediate postpartum placental auditing and documentation for any gross variability in size, shape, weight, completeness, anomalies, and tumors of trophoblastic and non-trophoblastic origins in addition to the scrutiny for secondary changes. The measurement of the umbilical cord length and inspection of the arteries, the vein, true or false knots and the Wharton's jelly should be routine and comprehensively recorded in a formatted protocol. All tissues have to be subjected for histopathological, immunological and histochemical studies as deemed necessary and facilities permit in order to understand and explain the underlying clinical disorder affecting the pregnancy and that of the mother's health [25-29].

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