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รายงานผู้ป่วย

A Case Report

Placental Hemangioma

Hemangioma ของรก

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ABSTRACT

Report of a pregnant woman, 29 years old, G4P3A0, was referred from Tayang Hospital to Prachomklao Hospital at 38 weeks gestation because of an abnormal finding in ultrasound that was suspicious for abnormal abdominal mass. Abdominal ultrasound a found placental mass and clinical diagnosis was placental hemangioma. Cesarean section was performed and found a big placental mass. The pathological diagnosis was placental hemangioma that was found in approximately 1% of pregnancies. Female baby weighs 3,390 grams, good Apgar score and no birth asphyxia.

บทคัดย่อ

รายงานผู้ป่วย 1 ราย เป็นผู้ป่วยหญิง อายุ 29 ปี G4P3A0 38 weeks gestation รับ refer มาจากโรงพยาบาลท่ายางด้วยเรื่องตรวจพบก้อนผิดปกติในช่องท้อง จึงขอส่งผู้ป่วยมาตรวจเพื่อยืนยันอีกครั้ง โดย abdominal ultrasound พบก้อนใน placenta. Clinical diagnosis คือ placental hemangioma หลังจากทำการผ่าตัดท้องคลอด พบก้อนขนาดใหญ่ในรก ผลทางพยาธิวิทยาเป็น placental hemangioma ซึ่งเป็นก้อนเนื้องอกที่พบน้อย ประมาณ 1% ของรกเท่านั้น ทารกเป็นเพศหญิง น้ำหนัก 3,390 กรัม Apgar score ดี ไม่พบภาวะขาดออกซิเจนขณะคลอด

Introduction

Hemangiomas of the placenta have long excited the interest of both pathologists and obstetricians and have been the recurring subject of several reviews (Siddall, 1926 ; Marchetti, 1939 ; de Costa, et al 1956 ; Strakosch, 1956 ; Fox, 1967 ; Philippe, et al 1969 ; Lin, et al 1970 ; Wallenburg, 1971 ; Rendina and Patrono, 1975 ; Sieracki, et al 1975 ; Schramm, et al 1987 ; Sfar, et al 1991 ; Tan & Yeo, 1992). They masquerade in the literature, however, under at least 22 synonyms, the only ones of which that are histogenetically correct, useful and

enjoy wide usage are 'chorangioma" and 'chorio-angioma'. This nomenclatural plethora is a reflection of the variable and pleomorphic histological appearances of the hemangioma, and there are no histogenetic or histological reasons for dignifying such a lesion in the placenta with a name that is separate and distinct from that given to it when occurring in other organs.[1]

The incidence has been reported between 0.2-139 : 10,000 births. Large tumors, those greater than 5 cm, have been reported to occur from 0.2-4 : 10,000 births. Smaller hemangiomas occur more frequently with and incidence of 14-139 : 10,000 deliveries. These smaller hemangiomas are often not diagnosed because they are not visible to the eye, and placentas are usually not examined by a pathologist. The overall accepted incidence in the literature is 1 in 100 births. The recurrence risk is not yet known but appears to be very small.[2]

A case report

History :

A pregnant woman, 29 years old, G4P3A0, 6 years old of last chid, was referred from Tayang Hospital to Prachomklao Hospital at 38 weeks gestational age because of abnormal abdominal mass on ultrasonogram.

Physical examination :

Vital signs: Temperature 36.8 C, Pulse rate 84/min, Respiratory rate 20/min, Blood pressure 130/70 mmHg

Abdominal examination : Height of fundus was $^{3}/_{4}$ above umbilicus, Fetal heart sound was 140/min, good fetal movement, No labor pain

Laboratory investigation :

Anti HIV : First time : Negative, Second time : Negative

VDRL : First time : Non-reactive, Second time : Non-reactive

Hematocrit : First time : 33%, Second time : 40%

Blood group : -B

Urine albumin 2+, Urine sugar : Negative

Ultrasound by gynecologist revealed a well circumscribed intraplacental mass with a complex echo pattern. Impression was placental hemangioma

Abdominal casarean section was performed and the placenta was sent the to Anatomical Pathological Division for examination. Female baby weighs 3,390 grams. Examination of the baby showed 49 cm in length and 39 cm each of head and chest circumferent. Apgar score were 9 (at 1 minute), 10 (at 5 minute) and 10 (at 10 minute). No birth asphyxia was present.

Pathological examination :

A placenta weighed 843.96 grams and measured 18x19x7 cm. The membrane was smooth, gray and glistening. The fetal and maternal surface were unremarkable. No evidence of infarction wasnoted. Serial sections showed a large well-defined rubbery dark brown mass, measuring 8 x 8 x 5 cm, covered by thin fibrous capsule. The umbilical cord measured 21.5 cm in length and 1.2 cm in diameter. It inserted at 4 cm from the margin. The cut surfaces showed two arteries and one vein.

Microscopic finding :

Sections of placental mass showed a mass composed of numerous normal size and dilated

capillaries, which were lined by mature endothelium and covered by fibrous capsule. Sections of umbilical cord showed two arteries and one vein. Sections of membrane were not remarkable change. No evidence of inflammation was noted.

Pathological diagnosis :

Placental hemangioma

Discussion

A hemangioma originates from primitive chorionic mesenchyme. It develops when blood vessels and stroma proliferate independently of the surrounding tissue. Marchetti describes three histological tumor types which are believed to represent various phases of tumor development.[3] One type is less differentiated or more immature with a compact structure of mostly cellular elements (Cellular type). The second type is the mature angiomatous or vascular type. This is the most common type of hemangioma composed of numerous small blood vessels and capillaries. The last type is characterized by degenerative changes (Degenerate type). Although tumors tend to be of one type, some may exhibit a combination of the characteristics described above.

Hemangiomas vary in size from a few millimeters to several centimeters in diameter. These tumors are usually a single mass but they can present as multiple separate masses. They are surrounded by a capsule or pseudocapsule. They usually located on the fetal surface of the placenta and are visualized as a mass bulging into the amniotic cavity. They can occur in the substance of the placenta or can protrude from the maternal side. Less often they may located in the membranes and attached to the placenta by a vascular pedicle. Hemangiomas have also been described on the umbilical cord. A variation of these findings has been described in a review article where the placenta was diffusely infiltrated by angiomatous tumor tissue.[4]

Placental hemangiomas show a very variable histological appearance (Angiomatous, Cellular and Degenerate) but should not be taken as indicating any fundamental difference between the various subgroups.

Mitotic figures may occasionally be seen in a placental hemangioma and very rarely, these are fairly numerous and associated with some degree of endothelial or stromal cell atypia. It has been suggested that tumors showing such features should be classified as 'atypical' hemangiomas and although this is a fairly meaningless term, its use is certainly preferable to classifying these neoplasms as sarcomas because there is no evidence that these tumors ever behave in a malignant fashion.

The first report of a hemangioma diagnosed by antepartum ultrasound was in 1978. Hemangioma is usually depicted as a well circumscribed intraplacental mass with a complex echo pattern. Uniform and nonuniform echogenic appearances and multicystic masses have also been representative of this tumor. The echo density of the well-delineated tumor differs from that of the placenta, allowing for its prompt recognition.

The vast majority of placental hemangiomas are of no clinical importance but a proportion are accompanied by a variety of complications that may affect the mother, the developing fetus or the neonate,

It has usually been maintained that these complications are only found in association with tumors measuring more than 5 cm in diameter. Large hemangiomas may cause severe complications such as fetal anemia, hydrops fetalis, fetal distress and fetal death.[5]

The most common clinical complication associated with hemangioma is hydramnios. The incidence of hydramnios has been found to related to the size of the tumor. It occurs in 18-35% of patients with large tumors.[6] A complication of hydramnios is an increased incidence of preterm labor, premature ruptured of membranes and preterm delivery.

Oligohydramnios has been reported to be associated with hemangioma. The diagnosis was made subjectively at the time of birth in a term gestation without the benefit of sonography. This association has not been confirmed by subsequent literature. There have also been reports of obstructed labor which were attributed to the size and location of the hemangioma. These reports have not been substantiated by more recent literature and appear to have been a coincidental rather than a causal finding.

The incidence of preeclampsia is believed to be increased by some[7] but others[8] believe the incidence is similar to that of the general population. Froehlich, using Collaborative Research Study data, has documented an increased incidence of preeclampsia of 16.4% vs 4.8% when comparing a group of 76 women with hemangioma to a control group of 44,994 women.

Various articles note the occurrence of antepartum and postpartum hemorrhage associated with hemangioma.[4,6,7] The antepartum bleeding is believed to be caused by a premature separation of the placenta as a result of bleeding from the tumor bed or a rupture of the vascular pedicle. Froehlich reported a 4% incidence of abruptio placenta in the group with hemangioma vs 1.2% in the control group. Postpartum hemorrhage has been reported to occur on occasions secondary to the over-distension of the uterus and subsequent uterine atony. Rarely the tumor has been reported to remain in the uterine cavity after delivery of the placenta and has caused postpartum hemorrhage.

Large hemangiomas have the potential to become arteriovenous shunts which can compromise the fetal circulation by increasing the venous return to the heart, thereby causing tachycardia, cardiomegaly and hypervolemia.[9] As a result, there is the possibility of high output cardiac failure, edema, hydrops and stillbirth.[10] Fetal anemia can also lead to hydrops through compensatory production of red cells by the liver, which causes hepatomegaly, portal hypertension and hepatic cell dysfunction, resulting in hypoproteinemia. The abnormal tortuous vascular channels in these tumors may cause red cell destruction and platelet sequestration, resulting in thrombocytopenia, microangiopathic hemolytic anemia, and disseminated intravascular coagulation. Feto-maternal hemorrhage may also cause fetal anemia. The abnormal vascular channels are non-functional placental tissue which do not allow for proper nutrient delivery and oxygenation of fetal blood. The decrease in functional placental tissue can lead to intrauterine growth retardation.[11]

In the older literature, hemangioma has been

associated with congenital anomalies but there has never been a cause and effect relationship established with any of these anomalies. An analysis of the Collaborative Research Study data reveals an increased incidence of various malformations in infants with intrauterine hemangioma when compared to controls.[7] This finding is probably due to a statistical aberration, since there is no valid reason for the supposed increased in malformations such as hip deformity, inguinal hernia, talipes, ear deformity, metatarsus deformity and pigmented nevus. Additionally, there is no connection between these deformities to allow for grouping them all with one condition. There does appear to be, however, a connection between hemangiomas and other vascular anomalies such as skin hemangiomas and single umbilical artery. The incidence of single umbilical artery in pregnancies complicated by hemangioma is 2.7% compared to 0.7% in the control group, and the incidence of skin hemangioma is 12.2% vs 2.1% in the control group.[7]

Pregnancies with hemangiomas have been noted to have a higher incidence of velamentous insertion of the cord (4.1% vs 1.5%). There have been reports of circumvallate placenta associated with hemangioma; however, the overall incidence appears to be the same as that in the general population. Placenta previa is also a condition that has been loosely associated with hemangioma but a true correlation has never been established.

Very little is known about the possibility of recurrence of placental haemangiomas in successive pregnancies.

Outcome is dependent on the size of the tumor. Smaller tumors have been found to be inconsequential. Large and diffuse tumors have been associated with pregnancy complications and therefore merit closer surveillance. A thorough ultrasound examination of the fetus should be performed to exclude any anomaly. Fetal karyotyping should be considered based on the few case reports of associated chromosomal abnormalities. In all cases serial sonograms should be performed to assess the growth of the tumor, growth of the fetus, and development of hydrops. During the third trimester, fetal well-being should be monitored with fetal movement counts, heart rate monitoring and biophysical profiles, as deemed necessary. The role of Doppler ultrasound for surveillance remains uncertain but should be performed to add to the information available and allow for future determination of its value in this setting.

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