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### RESEARCH ARTICLE

#### ANTENATAL DIAGNOSIS OF PLACENTAL CHORANGIOMA: ABOUT AN OBSERVATION.

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#### Abstract

Placental chorangioma is the most common non-trophoblastic placental tumor. The ultrasound allow antenatal diagnosis, but the diagnosis is confirmed the histopathological examination of the placenta after birth. It cause many maternal and fetal complications that are due to multiple intra-placental arteriovenous anastomoses. We report the case of a 25-year-old patient referred to our formation at the CHU Hassan II of Fez at 31 SA for a poorly tolerated polyhydramnios. Obstetrical ultrasound showed the presence of a 9cm subcutaneous chorionic mass with intra-lesional vascular flow suggestive of placental chorangioma. The fetus had subcutaneous infiltration associated with cardiomegaly. Cesarean section was decided after a course of corticotherapy. The newborn presented a favorable evolution with the appearance of multiple small hemangiomas scattered all over the body which spontaneously regressed. We have discussed through this observation the echographic characteristics of placental chorangioma, its maternal and fetal complications, and the fate of these pregnancies.

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#### Introduction:-

Placental chorangioma is the most common placental tumor with an estimated 1% (1). It is an intra-placental vascular proliferation most often of capillary type (2). These tumors act as large arteriovenous shunts in the placenta, diverting the blood from the fetus (3). The antenatal diagnosis is done by ultrasound coupled to the color Doppler (3). Ultrasonography reveals the existence of a placental tumor during routine ultrasound examination or during the pathological diagnosis of fetal complications (hydramnios, anasarca, fetal death etc.) (4) .

We report in this paper the case of a placental chorangioma diagnosed prenatally in our Obstetric Gynecology Department I at CHU Hassan II in Fez;

We have discussed through this observation the echographic characteristics of placental chorangioma, its maternal and fetal complications, and the fate of these pregnancies.

#### Observation:-

This is a 25-year-old patient, without significant pathological antecedent, G3P2, with two normal vaginal deliveries without incident. The patient was referred to obstetrics and gynecology obstetrics department of the CHU Hassan II

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of Fez at 31 SA for hydramnios of rapidly progressive installation. The patient presented, 15 days before her admission a significant increase in the abdominal volume hindering her mobility and the supine position.

Obstetrical ultrasound showed a mono-fetal pregnancy. The fetus was eutrophic and weight 2 kg. A hydramnios of great abundance was objectified defined by a large cistern at 15cm.

the ultrasound revealed the presence of a large placental chorionic mass of 9cm, well circumscribed, heterogeneous and having an oval form associated with placental thickening. Color Doppler showed intra-lesional vascular flow suggestive of placental chorangioma. The fetus had subcutaneous infiltration and a cardiomegaly. The rest of the morphological ultrasound was without particularity. The umbilical and cerebral doppler resistances were normal. The maximal systolic velocity of the middle cerebral artery was normal, without revelation of fetal anemia. An antenatal Corticosteroid Therapy for Fetal Maturation was administered. The therapeutic amniocentesis has been used to reduce polyhydramnios. The patient had a close clinical and ultrasound surveillance, and a fetal monitoring by the fetal heart rate recording. A caesarean delivery was decided at 32 weeks of aménorrhée.

At birth, the apgar of the female baby was 8 to 1 and 5 minutes of life respectively. The baby was pale and had a silverman score at 4/10. The baby's weight was 2300 g, a height of 43 cm and a cranial perimeter of 33 cm. The biological assessment showed anemia at 8 g / dl. The chest x-ray showed an aspect of stage 1 hyaline membrane disease. The baby was intubated and ventilated for 48 hours, transfused with red blood cell. The evolution was favorable after blood transfusion, extubated at the third day of life. Echocardiography showed a physiological dilatation of right ventricle and right atrium; associated to mitral and tricuspid valve regurgitation. A postnatal ultrasonographic screening of the urinary tract showed discreet bilateral pyelocalyceal dilation and normal size kidneys. On abdominal ultrasound: splenomegaly and absence of ascites. At the end of the first week, multiple small, diffuse hemangiomas appeared throughout the body. Abdominal ultrasonography and trans-fontanelle ultrasonography did not indicate deep localizations. The evolution was favorable with complete regression of hemangiomas.

Pathological examination of the placenta confirmed the diagnosis of placental chorangioma;

### **Discussion:-**

Placental chorangioma is the most common non-trophoblastic placental tumor with a frequency of 1 in 100 placentas (5). Only 50% are detected prenatally (5). It is found during careful anatomopathological examination in 1% of placentas (4). Its incidence increases with maternal age and is more common in multiple pregnancies (5). In our case, the patient was 25 years old, with a monofetal pregnancy.

The pathogenesis of placental chorangioma is not well known. Some authors believe that placental chorangioma results from a Pathological Placental Angiogenesis, Other consider that hypoxia and the decrease in arterial oxygen pressure is a contributing factor (6). Vascular endothelial growth factor (VEGF) as well as NEP and KIT play an important role in placental angiogenesis and the pathogenesis of placental chorangioma (7). Studies have shown that VEGF, NEP, and KIT are expressed in both myofibroblasts of neonatal cutaneous hemangiomas and in placental microcirculation, suggesting a common origin (7, 8).

The first ultrasound diagnosis of placental chorangioma was made by Asokan et al. 1978 (6). Ultrasound features make it possible to make the diagnosis in utero, but the confirmation of the diagnosis is histological by anatomopathological examination of the placenta after birth. Ultrasonographic diagnosis of placental chorangioma can be done by its two-dimensional ultrasound and Doppler characteristics, or by its fetal complications.

Placental chorangioma is characterized by an echogenicity similar to the placenta. It is an heterogeneous mass with sometimes hypoechogenic areas (corresponding to areas of necrosis). It has a rounded or oval shape and is located near the chorial plate often close to the insertion of the cord. A single umbilical artery can be associated with it. It is predominantly single mass but there are more rare forms of diffuse chorioangiomatosis. The antenatal diagnosis is possible if the size of the tumor exceeds 2cm (9). In our observation, the two-dimensional ultrasound had objectified a tumor mass well circumscribed mass having an oval form and a big size of 9 cm, echogenic, heterogeneous with hypoechoic areas.

At the color Doppler, the flow can be positive by the existence of intra-lesional arterial and venous vascularization (6). But this flow is not always found; This can be explained either by the fact that the tumor is composed mainly of small capillaries in which the speeds are too low to be detected, or by the evolution which can be marked by the occurrence of thromboses, necroses, fibroses or calcifications (6). We can also find signs of shunt with the umbilical artery. Using pulsed Doppler allows to find a high-speed arterial flow with resistance indices identical to those found in the umbilical artery (9). In our observation, the placental mass was very vascularized with the presence of intra-lesional vascular flow.

Fetal middle cerebral arterial peak systolic velocity is well correlated with fetal anemia which is a frequent complication of placental chorangioma (10). In our case, the maximum systole velocity was 1.1m/s and did not indicate fetal anemia.

Several fetal complications found on ultrasound should push the obstetrician to a good echographic study of the placenta not to miss the placental anomaly like placental chorangioma. These fetal complications are most often associated with large placental chorangiomas detectable on antenatal ultrasound. In our case, the ultrasound had objectified an important hydramnios, associated with a cardiomegaly and a subcutaneous and placental infiltration. IUGR and fetal anemia have not been objectified.

Placental chorangioma may pose a differential diagnosis problem with chorionic thrombosis, a plaque cyst, a deciduous hematoma, placental teratoma, aneurismal dilatation of a vessel under chorion, or placental infarction. In our case, the diagnosis of placental chorangioma was retained prenatally, by its pathognomonic ultrasound characteristics.

Positive diagnosis is confirmed by the histological examination of the placenta. Placental chorangioma is a vascular hamartoma consisting of a capillary network of mesenchymal chorionic origin. The hyperplastic vessels are encapsulated by a trophoblastic layer within the placental villi (7). This tumor can either be derived from the umbilical vessels, or be independent, pedunculated, separated by connective tissue (11). Its histological aspect is close to other cutaneous and visceral angiomas. The blood capillaries are anastomosed between them and sometimes connected to the umbilical vessels thereby deflecting part of the blood of the umbilical arteries to the benefit of the tumor and thus leading to functional placental insufficiency (6).

Many maternal and fetal complications can occur. These are due to the system of intra-placental arteriovenous anastomoses. The placental chorangioma is the seat of a shunt effect, responsible for a vascular flight and thus a fetal hypo-volemia. The risk of occurrence of complications is related to the importance of intra-lesional vascular shunts (4). Small placental chorangiomas are asymptomatic and do not complicate the course of pregnancy (12). Placental chorangiomas with significant clinical impact are tumors larger than 5 cm.

Maternal complications are metrorrhagia, preeclampsia, HRP, hemolytic anemia, maternal mirror syndrome with polyhydramnios and increased risk of premature labor and rupture of the placenta. In our observation, the hydramnios had caused a maternal gait during walking and lying down and led to a threat of premature delivery. Other fetal complications can be seen as prematurity, thrombocytopenia and fetal death in utero: There is a risk of sudden fetal death, probably due to massive fetal-maternal hemorrhage by rupture of vessels of the placental and L-shaped tumors. diffuse neonatal haemangiomatosis.

In our observation, fetal complications of placental chorangioma had led to premature labor at 32SA. One week after birth, multiple small hemangiomas diffuse all over the fetal body appear, without any other localization (normal liver ultrasound). Close prenatal surveillance should be instituted because of a decrease in the favorable outcome of pregnancies with placental chorangioma. (12) In the absence of fetal impact, regardless of the size of the tumor, close monitoring should be instituted, the rate of surveillance is adapted case by case according to the fetal tolerance. (6) It depends on the size of the chorangioma, its vascularity, the rate of growth, and the existence of complications. It should include (12) a clinical examination, cardiocathography, echographic and velocimetric. Ultrasound surveillance will look for fetal cardiomegaly, tricuspid insufficiency, hepatomegaly, dilation of the umbilical vein, umbilical artery Doppler abnormality and Arantius canal, an evaluation of the amount of amniotic fluid. Heart failure or fetal anemia prior to installation of the fetus-placental anasarca. In our case, the patient was seen at an advanced stage at 32SA. Fetal complications (cardiomegaly, hydramnios with placental) required fetal

extraction. Fetal surveillance only lasted 2 days for the administration of corticosteroids for pulmonary maturation and one fetal heart rate recording two times per day.

Vaginal delivery is not contraindicated; but most often a cesarean is performed because of the maternal and fetal complications, as is our case (prematurity, heart failure, anasarca ...) .If complications occur at an advanced age of pregnancy, delivery should be discussed according to fetal maturation (12). But if the complications occur in the 2nd trimester, or the extraction can not be considered because of the prematurity, different therapeutic modalities must be discussed. The objective of the term of the delivery after a laser treatment is of 34 SA (4) .

The symptomatic treatment is amniodrainage in case of hydramnios important and poorly supported. It (10) improves maternal tolerance and decreases the risk of obstetric complications associated with hydramnios (4). In our observation, the patient benefited from an amniodrainage to relieve the maternal gene. Transfusion in utero is the second symptomatic treatment frequently found in the literature. Uterine transfusion may be used for antenatal diagnosis of fetal anemia with mean cerebral arterial PVC greater than 1.5 MoM. But there may be true fetal anemia in placental chorangioma. It is most often a hypo-volemia. The transfusion could then cause fetal heart decompensation. Some authors recommend that transfusion in utero be reserved for post fetoscopic fetal-maternal haemorrhage (4).

New interventions have been proposed to block vascular cancellations of the tumor with limited success in the majority of cases (12): Absolute alcohol injection, endoscopic laser coagulation and other techniques such as endoscopic sutures by bipolar, electro surgery have been presented in some cases of literature without reported survival. (12) The treatment of choice is laser coagulation of anastomoses by fetoscopy. It only requires local anesthesia and does not require precise placement of the needle in a supply vessel as in the use of toxic substances such as alcohol injection (12). This treatment is the most effective in reducing fetal heart burden and anasarca without associated toxicity. The main complication is fetal-maternal haemorrhage with acute fetal anemia. (4) The prognosis of placental chorangioma depends on the characteristics of the placental chorangioma and on the occurrence of maternal and fetal complications. The size of the chorangioma and the early diagnosis are prognostic factors. Choroangiomas greater than 4 cm are associated with 30% perinatal mortality, (10). For most authors, tumors less than 4 cm are usually of good prognosis. The size of the tumor can quickly evolve, thus constituting an element of bad prognosis. The placental chorangioma can exceptionally regress spontaneously. A case has been reported in the infarction literature of placental chorangioma with regression of fetal anasarca. (6) Color Doppler can also help to determine the prognosis. It allows to estimate the degree of vascularization of the placental tumor. The less vascularized the tumor is, better is the prognosis, due to the decrease in feto-tumoral vascular shunts (13). An intense vascularization of the placental mass constitutes a higher risk of fetal complications. Doppler ultrasound with fetal echocardiography can also diagnose fetal heart failure. In our case, color Doppler showed a highly vascular tumor with intra-lesional arterial flow causing several complications. Fetal complications are also an element of poor prognosis. Heart failure and foeto-placental anasarca are the most serious complications feared. The existence of fetal cardiomegaly preceding fetal anasarca is a criterion for fetal extraction. (3). Fetuses who develop anasarca are most at risk for perinatal mortality (3). Other fetal complications have an impact on perinatal morbi mortality such as IUGR with chronic fetal distress, anemia and fetal thrombocytopenia and premature delivery.

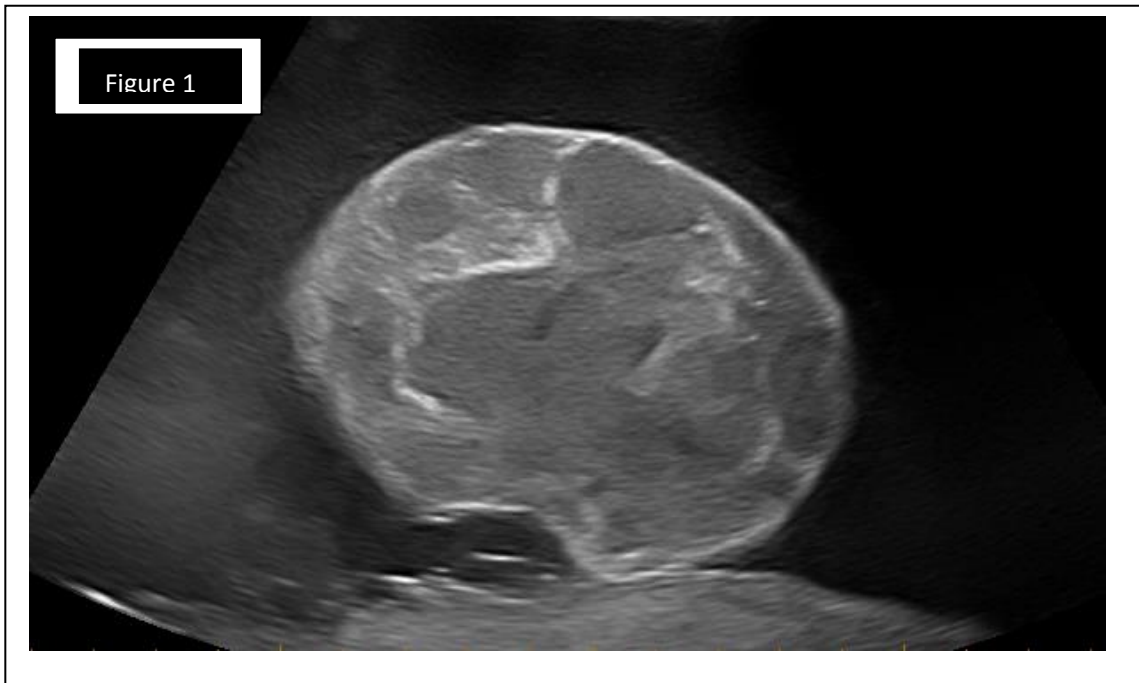
### **Conclusion:-**

Placental chorangioma is a rare but serious pathology whose antenatal ultrasound is the cornerstone for diagnosis and surveillance in order to hope for a favorable outcome of these pregnancies.

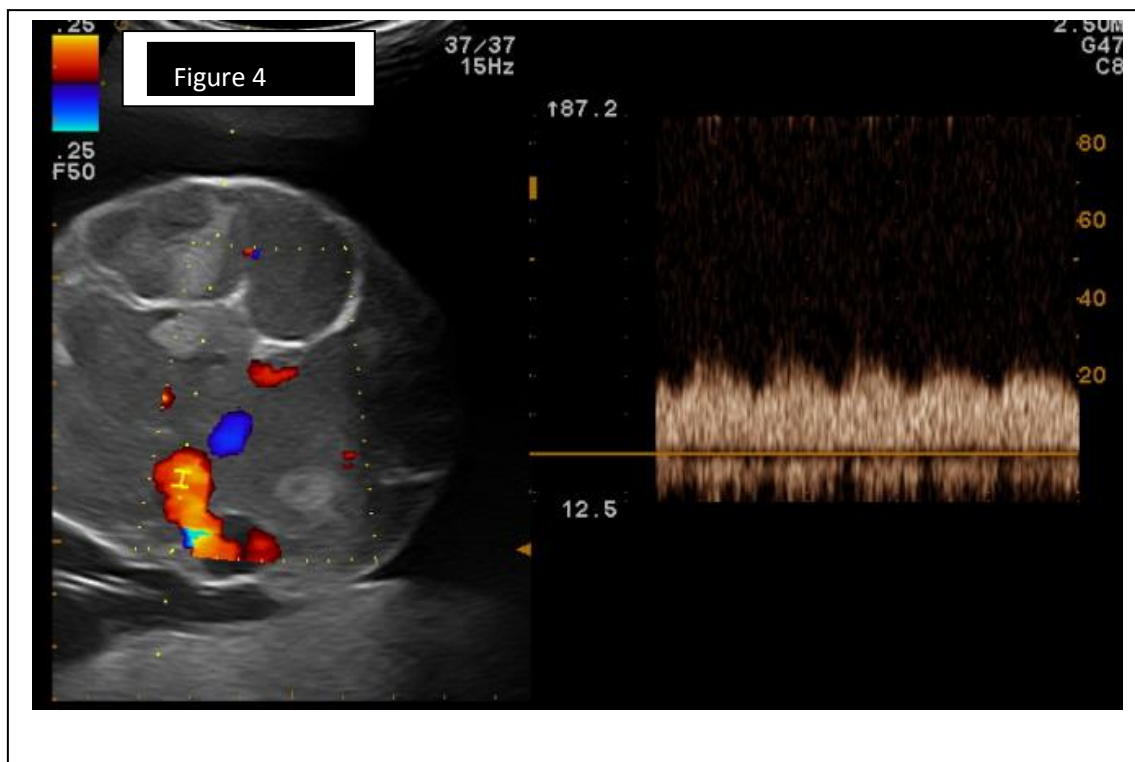
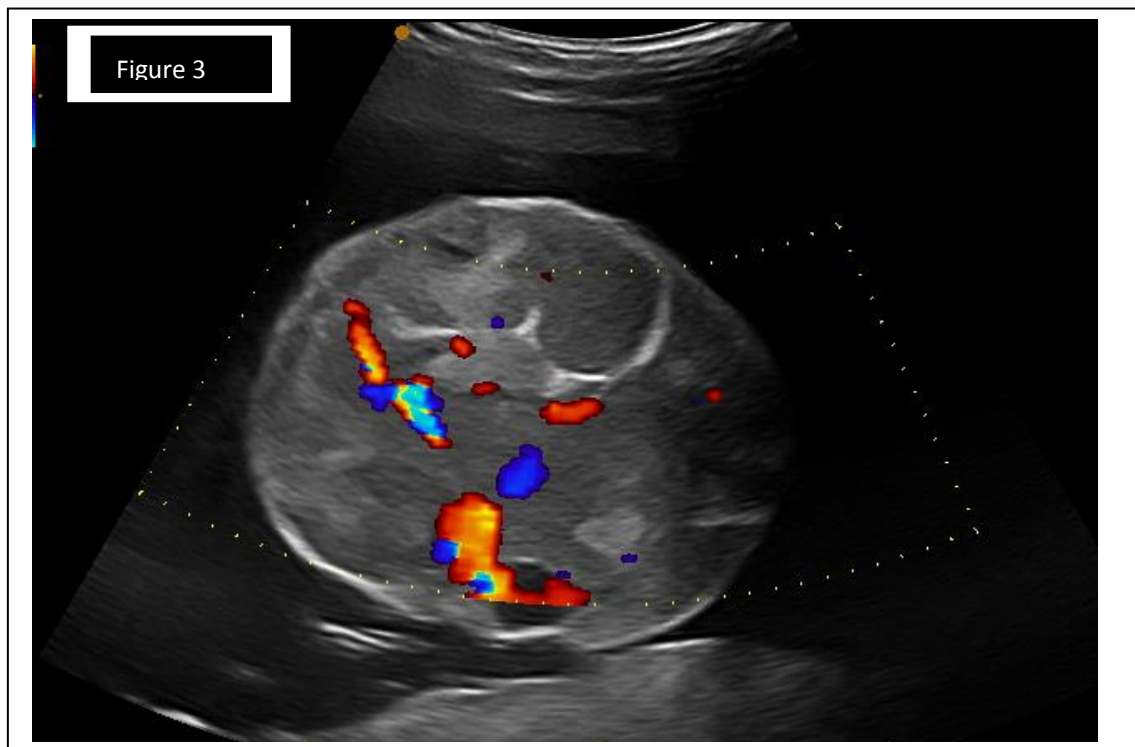
It allows positive diagnosis by its characteristics in two-dimensional mode and Doppler mode, and by the detection of fetal complications. Ultrasound also has a role in differential diagnosis, pregnancy monitoring and predicting neonatal prognosis.

Placental chorangioma should be routinely investigated in the presence of fetal hydramnios, heart failure, IUGR, placental fecal anasarca, or unexplained fetal death to initiate an appropriate therapeutic approach early.

**Figure 1 and 2:-**well-circumscribed tumor mass of 91x68 mm, oval-shaped, echogenicity similar to that of the placenta, heterogeneous in favor of a placental chorangioma with 52 mm placental thickening

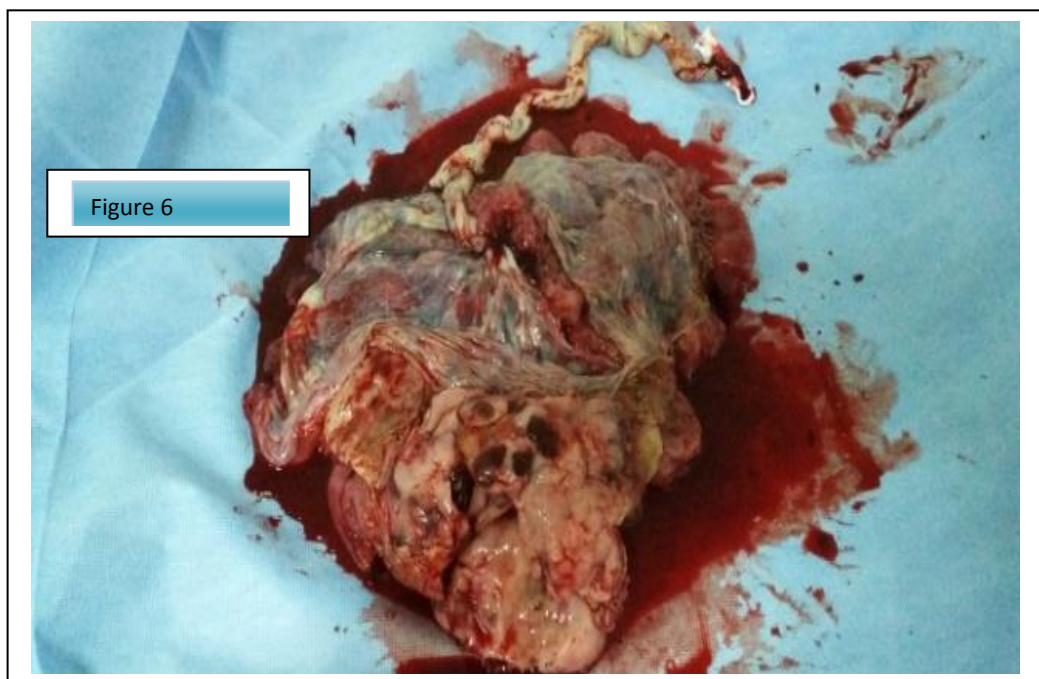


**Figure 3 and 4:-Color Doppler: Intra-lesional vascular flow**





**Figure 5 and 6:-macroscopic image of the placenta**



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