Case report

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A Rare Case of Chorioangioma / Non Immune Fetal Hydrops

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Abstract

Chorioangioma is a placental hemangioma is a common, non-trophoblastic benign vascular placental tumour of primitive chorionic mesenchyme. The size of the tumour is important. Smaller tumours are clinically insignificant. Giant chorioangioma more than 4 cm has higher risk of maternal and foetal complications. Early diagnosis is done by imaging techniques. Placental lesions detected on sonography necessitate close surveillance of these pregnancies because of the poor outcome ofpregnancy. We present a 32-year primigravida with placental chorioangioma who went in spontaneous labour and delivered a male baby. The baby was referred to higher centre for further evaluation and revealed mild cardiomegaly. The histopathological examination of placenta helped in the diagnosis of placental chorioangioma.

Keywords: Hemangioma; Chorioangioma; Placental; Maternal and foetal complications.

INTRODUCTION

The term non--immune hydrops fetalis (NIHF) is defined as an edematous foetus with fluid collections in some or all serous cavities that does not have erythroblastosis fetalis from isoimmunisation. On immune fetal hydrops was first described in 1943 by Dr Edith Potter[1]; at present more than 80 conditions are known to be associated with hydrops with very high perinatal mortality ranging from structural heart disease, fetal arrhythmias, chromosomal anomalies, intrauterine infections and larger chorioangiomas of the placenta.[2–5] In most countries with low rhesus negative rates in the population, non-immune causes are more prevalent; the incidence depends on the region and also varies seasonally in relation to parvovirus B19 epidemics. Chronic placental insufficiency is the commonest cause for fetal growth restriction. Rare placental causes affecting fetal outcome are partial mole, chorioangioma, and placental teratoma. Large chorioangioma has adverse effects on both mother and foetus. We report a huge chorioangioma resulting in polyhydramnios, preterm labour.

CASE REPORT

A 32-year-old G2P1L1 at gestational age of 32 weeks + 5 days with previous normal delivery with known case of bronchial asthma came with complaints of abdominal pain and abdominal distention for 4 days, reduced perception of fetal movements for 2 days was admitted and evaluated. Ultrasound obstetrics done and was suggestive

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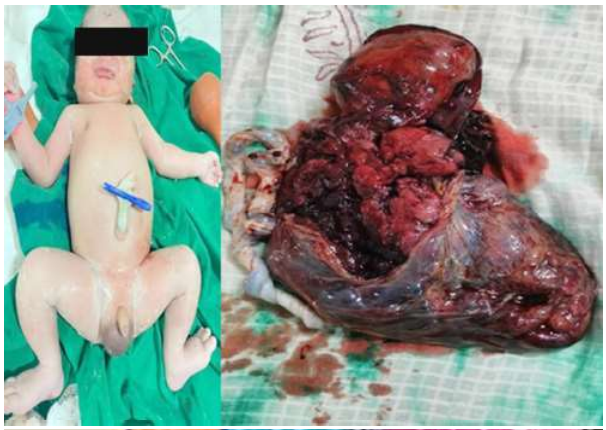
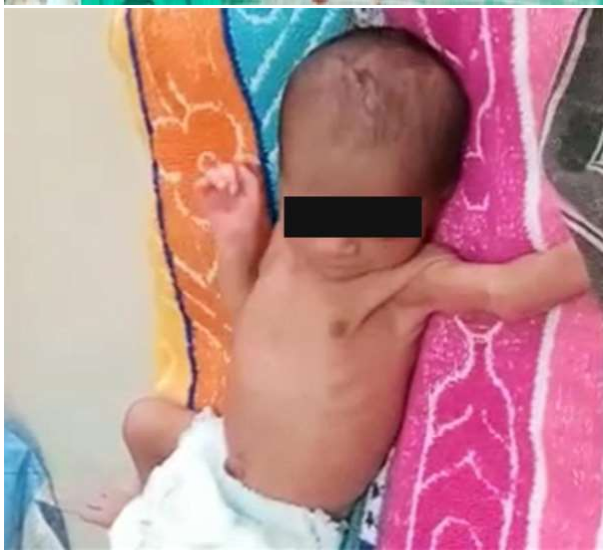
of polyhydramnios (AFI-29 cms), diffuse scalp edema, subcutaneous edema of foetus. Mild fetal enlargement of right atrium and right ventricle with tricuspid regurgitation. No ascites, pleural effusion. Hypoechoic lesion of 6.96 x 4.51 cms seen in the right side of placenta and Doppler study suggestive of mild peripheral vascularity suggestive of chorioangioma. Blood sugar values and MCA Doppler were normal. Patient diagnosed with acute polyhydramnios with non-immune fetal hydrops. Patient spontaneously progressed to active stage of labour and delivered an alive male baby of birth weight 2.48 kg with APGAR.[8,9,10] Baby showed features of scalp edema, periorbital edema and ascites. Baby was referred to higher centre for further evaluation, all investigation was done and found normal. USG suggestive of mild cardiomegaly. Baby got discharged, alive and healthy now.





DISCUSSION

Placental chorioangioma is the most common benign tumour of the placenta. They were more seen in multiple pregnancies and in female babies. Chorioangioma is believed to arise by 16th day of fertilization, although there is no documentation of tumour in first trimester. It consists of a benign angioma arising from chorionic tissue. Three histological patterns of chorioangiomas have been described by Marchetti-angiomatous, cellular, and degenerate. The angiomatous is the most common, with numerous small areas of endothelial tissue, capillaries, and blood vessels surrounded by placental stroma. These lesions are some times classified as placental hamartomas rather than true neoplasia. There is no malignant potential. They are benign lesions with no malignant potential. Recurrence of chorioangioma in subsequent pregnancies is rare. They're usually single, small but may present as multiple placental masses of varying sizes or giant tumours measuring > 5cm. In cases of small tumours, the fetus is at low risk and the pregnancy proceeds normally. Large tumours are frequently associated with maternal and fetal morbidity, which includes polyhydramnios, due to transudation of fluid caused by the tumour causing mechanical obstruction of blood flow near the cord insertion or placental insufficiency due to shunt mechanism of tumour vasculature. Another striking but rare complication associated is non - immune hydrops fetalis. Large chorioangioma contains vessels which act like arteriovenous shunts causing shunting of blood which alters fetal hemodynamic leading to non-immune hydrops. On grey scale ultrasound, chorioangioma is a hypo/hyperechoic well-defined mass, classically seen protruding into the amniotic cavity from the fetal surface near the umbilical cord insertion. It is distinct from the placenta, and contains anechoic cystic areas which corresponds to blood vessels or necrotic areas. Spectral-Doppler adds other data that leads to the diagnosis; it demonstrates the same heart rate as the fetus helping to confirm the connection of the mass to the fetal vasculature[1], also differentiating it from other placental tumours like partial hydatidiform mole, teratoma, placental hematoma and uterine submucosal fibroid, which share some of the imaging features. MRI shows a heterogeneous mass with high T2 signal in comparison to the placenta. T1W sequences showed iso/hypo intense mass, sometimes with peripheral hyper intensity related to hemorrhage. When acute infarct/degeneration occurs, this may improve the fetal prognosis but, on the other hand, may lead to potential mistakes, such as placental haematoma diagnosis due to lack of demonstrable blood flow. Asokan *et al.* in 1978 made the first sonographic diagnosis of placental

chorioangioma.[9] USG, which is routinely done, has no specific diagnostic finding. However, in some cases, it is seen as a well circumscribed lesion with the typical appearance of a vascularized tumour different echogenicity chogenicity as compared to the normal placental tissue. Large tumours can be of variable shapes. These GCs are on the fetal surface near the umbilical cord insertion site protruding in the amniotic cavity. Chorioangiomas are supplied by fetal circulation. It acts as a physiological dead space returning oxygen depleted blood to the fetus, resulting in chronic hypoxia leading to fetal growth restriction. The pathological changes seen in the neonates are anaemia, thrombocytopenia, oedema, non-immune hydrops fetalis, still birth, prematurity, intrauterine growth retardation, or fetal death. Arteriovenous shunts in GC cause impairment of the fetal circulation. There is an increase in the venous return to the heart which leads to hypervolemia, tachycardia, cardiomegaly, and congestive heart failure. The fetal red cells can be traumatized while traversing the labyrinthine of newly formed and deformed vascular channels.

This can be a cause of micro angiopathic hemolytic anemia. Sequestration of blood in the vascular mass can also be the reason for anemia and thrombocytopenia.[1,8] On USG, chorioangioma has to be differentiated from other lesions. These lesions include sub chorionic cyst which is located near the umbilical cord insertion. Hematoma and teratoma of the placenta, degenerating myoma, and deceased twin are the other conditions which can be differentiated using color Doppler or magnetic resonance imaging in most cases. Angiomatous, cellular, and degenerate are the three histological patterns of chorioangioma. The common being angiomatous pattern. It shows proliferation of blood vessels. These vessels are in different stages of differentiation. This leads to capillary or cavernous vascular spaces having placental stroma. The tumour cells show strong immuno reactivity for CD31, CD34, factor-VIII, GLUT 1, and cytokeratin.[18] The positivity of these immuno markers suggests that the tumour arises from the blood vessels of the chorionic plate and anchoring villi.[5] Histologically, chorangioma has to be differentiated from chorangiosis and chorangiomatosis. In these two conditions, there is diffuse or more often focal villous angioblastemal proliferation. In chorioangioma, the proliferation is not within the villi. "Cholangio carcinoma"a probable misnomer shows trophoblastic proliferation in the vicinity of the chorioangioma is the other condition which needs to be differentiated.

Intervention

Chorioangioma with complications before fetal viability requires interventions. Ultra sound along with colour Doppler study provides early comprehensive information of the tumour as well as fetal well being. MRI can be used for further evaluation in case of equivocal ultrasound findings, hence appropriate, timely intervention can be planned, reducing maternal and child morbidity. Polyhydramnios is treated with maternal indomethacin therapy. Steroid administration for acceleration of fetal lung maturity before 34 weeks is indicated.

CONCLUSION

Early diagnosis and timely intervention will save the baby and reduce neonatal complications. Placental chorioangiomas of large size are rare and the prognosis is poor when a big tumour causes fetal hemodynamic changes with NIHF, but treatment of heart failure may be promising in these new borns and complete recovery is achieved in some cases.

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