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RESEARCH ARTICLE

PLACENTAL CHORANGIOMA AND ITS CLICOPATHOLOGICAL SPECTRUM: A CASE SERIES

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Abstract

Introduction: Chorangioma is a rare primary vascular tumour of placenta with an incidence of about 1% in carefully examined term placenta. Chorangiomas in the past have been described as hemangiomas. Chorangioma is a benign, vascular, non-trophoblastic tumor of plancenta that is potentially threatening and can lead to hazardous feto-maternal catastrophy.

Objectives: To analyse clinicopathological characteristics, diagnosis, differential diagnoses, treatment, and prognoses of placental chorioangioma (PCA).

Results: Mean age of the patients was 38 years. 2 patients were below 30 years and 2 were above 30 years. Only 1 patient was primigravida, rest 3 were multigravida. Mean gestational age was 31.8 weeks. 3 out of 4 cases were term gestations. Out of 4 patients, 3 had an emergency LSCS and 1 had normal labor. 1 out of the 4 patients delivered a dead fetus. Grossly, all the specimens werecomplete placenta. All lesions were extraplacental, solitary and all were >5cm. The mean tumor size was 7.3 cm (range: 6-8.5cm). All the tumowere nodular, well circumscribed, solid masses. On cut-section, lesions were reddish-brown to tan-off-white, solid, firm, hemorrhagic masses as depicted in the pictures. Microscopically, the lesions were composed of network of proliferating capillaries lined by benign endothelium in enlarged villi covered by trophoblastic epithelium. Focal hyalinized areas were seen in one of the cases as depicted in the pictures.

Conclusion: Giant chorangiomas are relatively rare, however, their diagnosis and management pose a big challenge owing to serious antenatal feto-maternal complications they lead to. Careful radiological and histopathological examination are thus of utmost importance.

Introduction:-

Chorangioma is a non-trophoblastic benign vascular tumour of the placenta. It arises from major stem villi under the chorionic plate or at the placental margin.Chorangiomais a rare primary vascular tumour of placenta with an incidence of about 1% in carefully examined term placenta.(1) It is considered hamartoma-like or hyperplastic capillary lesion rather than a true neoplasm.Chorangioma,although, is the most common benign tumor of placenta (2) but tumors arising from placenta are as such rare.Elderly primipara, twin pregnancies, hypertension,diabetes and

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high altitude are often associated with chorangiomas.(3) The exact etiology however is not known. The clinical significance of chorangioma is related to its size. Small chorangiomasare often asymptomatic and an incidental finding. They tend to regress spontaneously. On the contrary, giant chorangiomas>5 cm in diameter are rareand clinically significant as they are often associated with a variety ofmaternal and fetal complications(4) likepolyhydramnios,toxemia,preterm placenta separation,placenta previa,preterm labor, AV shunts etc.and a poor perinatal outcome.Diagnosis of large chorangiomas can be made in the antenatal period by either colourdoppler or ultrasonogram.(5)

Material And Methods:-

This study was conducted in the Department of Pathology of a tertiary care hospital in Jammu (J&K). Placenta samples were studied over a period of 3 years, from May 2016 to May2019. A total of 4 cases of placental chorangiomas had been reported during this period. Case details like mother's age,order of pregnancy, gestational age, mode of delivery, indications for delivery, pregnancy outcome and birth weight of the children were obtained. Further,details of relevant investigations, procedure done, nature and gross features of the specimens and histopathological diagnosis of the tissues were obtained.

Haematoxylin and Eosin stained sections of the paraffin blocks of the representative cases were reviewed.

Inclusion Criteria:
All placental specimens with mass were included for the study.

Exclusion Criteria:
Placental specimens received for other pathological conditions were excluded from the study.

Aim And Objective:-

To look at the frequency distribution of clinical parameters of the cases, fetal outcome, and the gross and microscopic features of the placenta submitted for histopathological examination.

Results:-

A total of 4 cases of chorangioma were identified in the duration of 3 years. Mean age of the patients was 38 years (range: 24-45 years). 2 patients were below 30 years and 2 were above 30 years. Only 1 patient was primigravida, rest 3 were multigravida. Mean gestational age was 31.8 weeks (24-38 weeks). 3 out of 4 cases were term gestations (>28 weeks). Out of 4 cases 3 were delivered off by emergency LSCS and 1 was delivered off by normal labor. The indications for emergency LSCS were fetal distress (in 1st case), abruption placenta (in 3rd case) and placenta previa (in 4th case). Pregnancy outcome was mostly good with alive and healthy babies in 2 cases, 1 patient delivered an alive baby with IUGR and another patient delivered a dead fetus. Birth weight was low in 1 case and very low in another one. Grossly, all the specimens werecomplete placenta. All lesions were extraplacental, solitary and all were >5cm. The mean tumor size was 7.3 cm (range: 6-8.5cm). All the tumors were nodular, well circumscribed, solid masses. On cut-section, lesions were reddish-brown to tan-off-white, solid, firm, hemorrhagic masses as depicted in the pictures. Microscopically, the lesions were composed of network of proliferating capillaries lined by benign endothelium in enlarged villi covered by trophoblastic epithelium. Focal hyalinized areas were seen in one of the cases as depicted in the pictures.

Case no.	Age (years)	Gravida	Gestational age (weeks)	Mode of delivery	Indication	Delivery outcome	Birth weight (kg)
1	24	1	24	Emergency LSCS	Fetal distress	Dead born	0.9
2	35	2	28	Normal vaginal delivery	Active labor	Healthy and alive	3
3	45	4	35	Emergency LSCS	Abruption placenta	Healthy and alive	2.6
4	30	3	30	Emergency	Placenta	IUGR but	

				LSCS	previa	alive	

Table 1:- Showing the clinical details of the 4 patients.

Case no.	Specimen type	Size of the lesion	No. of lesions	Cut surface
1	Placenta with an extraplacental mass	8cm	1	Solid, reddish-brown, Hemorrhagic
2	Placenta with an extraplacental mass	6cm	1	Reddish brown, Hemorrhagic
3	Placenta with an extraplacental mass	7cm	1	Tan-off white, focal blood-filled spaces
4	Placenta with an extraplacental mass	8.5cm	1	Firm,reddish-brown Hemorrhagic

Table 2:- showing the morphological details of the placentas.

Figure 1:- Gross picture showing extraplacentalchorangioma.

Figure 2:- Gross picture showing cut surface of chorangioma.

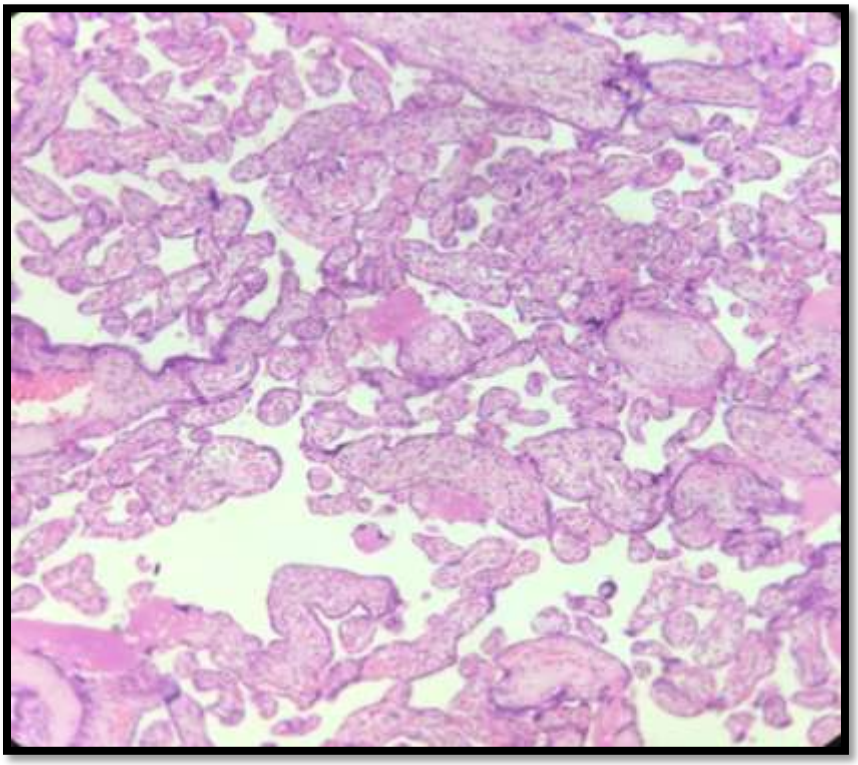

Figure 3:- Low power view of chorangioma showing varying sized chorionic villi, H&E x100.

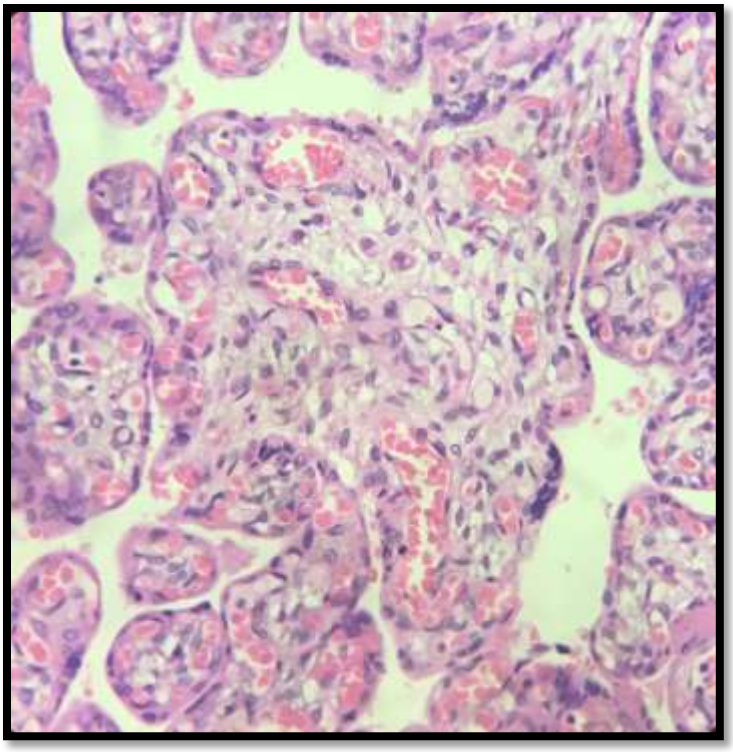

Figure 4:- High power view of chorangioma showing highly vascularized chorionic villi, H&E x400.

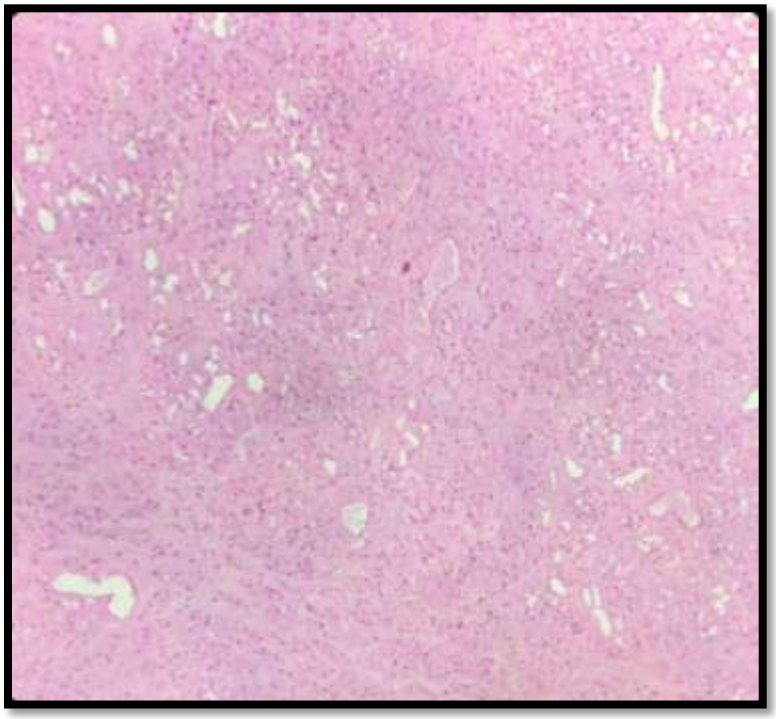

Figure 5:- Network of capillaries and sinusoids, H&E x40.

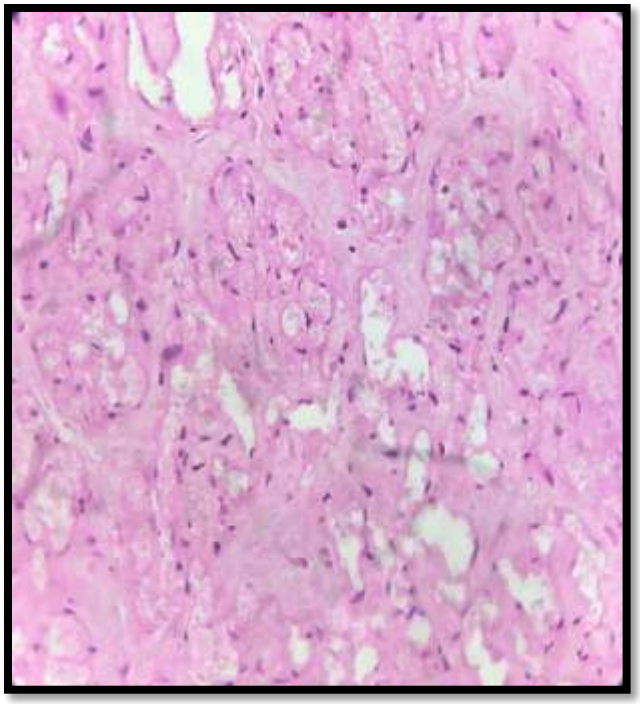

Figure 6:- Lobules of congested capillaries, H&E x100.

Discussion:-

Chorangiomas in the past have been described as hemangiomas. Placental angioma, chorionangioma, angiomyxoma or vascular harmatoma of the placenta are the other names given to placental chorangioma.(6) Chorangioma is a benign,vascular,non-trophoblastic tumor of plancenta that is potentially threatening and can lead to hazardous feto-maternal catastrophy. Its prevalence is 1%, when placenta specimens are grossed and explored carefully.(7) Exact etiology is not known,however they are reported to be arising from the stem villi.Depending upon size and number, Ogino and Redline classified villous capillary lesions of placenta into chorioangioma, chorioangiomatosis and chorioangiosis.(8)Ultrastructurally,chorioangioma and chorioangiomatosis tend to be lesions of mature stem villiwhereaschorioangiosis tends to be a lesion of terminal villi. Many variations of chorangioma histology are recognized and include capillary, cavernous, endotheliomatous, fibrosing and fibromatous forms. Furthermore, Marchetti described three patterns of chorioangiomas: angiomatous, cellular and degenerated.(9) Angiomatous is the most recurrent pattern which is composedof numerous small areas of endothelial tissue, capillaries, and blood vessels surrounded by placental stroma. These lesions are thus classified as placental hamartomasrather than true neoplasia and do not possess any malignant potential.However, a chorangioma can sometimes be cellular and harbor significant mitotic activity that raises the possibility of a malignancy.In literature, a few reports of mitotically active chorangiomas are present. Some authors have reported increased mitotic activity and cellularity in this rare tumor and have suggested that these occasional chorangiomas might represent sarcomas. However, true invasion and metastasis were never demonstrated in these cases. Moreover,there has been no convincing report of a chorangioma with malignant clinical behavior. (10) The known general agreement in literature about the association of larger chorangiomas withadverse feto-maternal clinical complications was seen in the present study also. All the 4 chorangiomasidentified in the present study were >5cm and were associated with feto-maternal complications like polyhydramnios, placenta previa, placenta abruption and fetal distress. Similar results were seen in a study by Balamurugan V et al. (11) They conducted a study on placental specimens over a period of 7 years which yielded 7 chorangiomas,all of which measured >5cm, with complications like abruptio placenta with cord presentation, acardiac twin with placental solid tumour, fetal distress and atonic post-partum hemorrhage. Other common complications of chorangiomas are fetal anemia, thrombocytopenia, nonimmunehydrops, fetal heart failure, fetal cardiomegaly, fetal growth restriction and preterm delivery.(12)Although the underlying pathophysiology for these complications has not been clearly illustrated, it is believed that arteriovenous shunts, sequestration of red blood cells and platelets by tumor play an important role.(13-17) All this leads to hemolysis, microangiopathic anemia, and reduced oxygen carrying capacity of blood which results inhydropsfetalis.Feto-maternal hemorrhage across tumor capillaries leads to elevated AFP levels in maternal blood and ultimately maternalhemolysis. Polyhydramnios has

been accredited to surplus amniotic fluid transudateflowing through walls of abnormal tumor vessels, as well as to mechanical obstruction of umbilical vein by large tumor mass.(18,19)Preterm delivery,as seen in 3 cases in the present study,is a common after-effect of hydraminos.(20)Chorangioma often results in IUGR, which was seen in case number 4 in the present study. Chorangiomaserves as a physiological dead space, giving rise to chronic hypoxia resulting in fetal growth restraint.Radiologically, chorangioma often presents as a well-defined complex echogenic mass which appears to be different from the rest of placenta. It often extrudes into amniotic cavity near umbilical cord insertion. Color doppler imaging is often used for confirmation of diagnosis of placental chorangiomas. It has made detection of vascularity of the tumor and angiomatous, cellular and degenerative changes in chorangiomas much more effortless.(21) Placental MRI is also being used nowadaysaccessory to ultrasound placental imaging.(22)Grossly, chorangiomas are usually well circumscribed, nodular masses found on the fetal surface of placenta near the insertion of the umbilical cord. Sometimes they are found within the placental parenchyma. The cut surface is variable with solid, firm, brown, yellow, tan,purple, red or white masses.Secondary changes like myxoid areas,hyalinization and calcification can be seen.(23)Microscopicallychorangiomas are well circumscribed, nodular tumors, exhibitingproliferation of capillary sized vessels resulting in expansion of contiguous affected villi. Chorangiomas are mainly composed of a mixture of endothelial cells, pericytes and myofibroblasticstromal cells.Associated benign, nonspecific surface trophoblastic proliferation is also seen in up to 40% cases. Occasionally degenerative changes like hyalinization, necrosis and calcification are present.

Immunohistochemical stains like CD31, CD34, factor VIII antigen and GLUT1positively stain the endothelial cells and CD18 suggests origin from chorionic plate and anchoring villi.Chorangiomas must be differentiated from other villous capillary lesions, namely chorangiomatosis and chorangiosis.Chorangiosis (villous vascular proliferation) is a vascular lesion involving the terminal chorionic villi. In normal placenta, chorionic villi rarely contain more than 5 vascular channels.However,chorangiosisas described by Altshuler in 1984, is the presence of a minimum of 10 terminal villi, containing more than 10 capillaries per villus in 10 medium power fields in at least 3 or more random, noninfarcted placental areas. (24) Also in chorangiosis,individual capillaries are surrounded by distinct basement membranes and there is no circumferential lining of pericytes or loose reticulin bundles. On the other hand, chorangiomatosisis a nonexpansile vascular proliferation similar tochorangioma occurring in otherwise normal stem villi.Grossly,the lesiondoes not form a discrete mass. Microscopically,the lesion permeates normal villi and is composed of proliferation of small capillaries surrounded by a continuous thick layer of pericytes which stain positive for smooth muscle actin (SMA). Treatment modalities are either supportive or definitive. Supportive measures include serial fetal intrauterine transfusions for fetal anemia, and amnioreduction through therapeutic amniocentesis/maternal transplacental pharmacotherapy with indomethacin. (25,26,27) Steroid administration is helpful in accelerating fetal lung maturity before 34 weeks. Definitive endoscopic surgical devascularisation (ligation/clipping, fetoscopic laser ablation, embolization, chemosclerosis with absolute alcohol injection) and radiofrequency ablation of tumor vessels are also helpful.(28)

Conclusion:-

Giant chorangiomas are relatively rare, however, their diagnosis and management pose a big challenge owing to serious antenatal feto-maternal complications they lead to. To prevent the adverse pregnancy outcomes early diagnosis, individual assessement and institutional delivery is of utmost importance. Overlooking of these lesions, thus, is to be prevented by careful radiological and histopathological examination.Interventions such as endoscopic laser coagulation, radiofrequency ablation of tumor vessels etc. may be useful for selected patients on regular fetal monitoring to avoid preterm delivery in women placental chorangiomasof larger size (>5cm).

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