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ONLINE CASE REPORT

Placental Tumour
What could it be?

*Nihal Al-Riyami,[1] Rahma Al-Hadabi,[1] Tamima Al-Dughaishi,[1] Marwa Al-Riyami,[2]

ورم المشيمة
ماذا يمكن أن يكون؟

نهال الريامي، رحمه الهدابي، مروة الريامي

الملخص: **تشمل أورام المشيمة على أورام وعائية مشيمية، أورام مسخية، أورام وعائية دموية وأورام دموية. الأورام الوعائية المشيمية هي أورام دموية حميدة وأكثر أورام المشيمة شيوعاً، بواعق انتشار 1%. الأورام الوعائية المشيمية الكبيرة تعتبر نادرة وقد تؤدي إلى مضاعفات الحمل وسوء نتائج الفترة المحيطة بالولادة. وتشمل هذه المضاعفات فقر الدم الجنيني، استسقاء الجنين، تقييد نمو الجنين، موه السلى والولادة قبل الأوان. نعرض هنا تقرير حالة ورم وعائي مشيمي كبير، وإدارة الرعاية السابقة للولادة، ونتائج الأمومة والجنين.**

مفتاح الكلمات: المشيمة؛ ورم وعائي مشيمي؛ فقر الدم؛ الجنين؛ موه السلى؛ تقرير حالة؛ عُمان.

ABSTRACT: Placental tumours include placental chorioangiomas, teratomas, haemangiomas, and haematomas. Placental chorioangiomas are benign vascular tumours and are the most common placental tumours, with a prevalence of 1%. Large placental chorioangiomas are rare and may lead to pregnancy complications and poor perinatal outcomes. These complications include fetal anaemia, *hydrops fetalis*, fetal growth restriction, polyhydramnios, and preterm delivery. We report a case of a large placental chorioangioma, the antenatal management and the maternal and fetal outcomes.

Keywords: Placenta; Chorioangioma; Anemia, fetal; Polyhydramnios; Case Report; Oman.

Placental tumours are broadly divided into trophoblastic and non-trophoblastic tumours. The latter include chorioangiomas, teratomas, haemangiomas and haematomas. Placental chorioangiomas are benign vascular tumours and are the most common placental tumours with a prevalence of 1%.[1] Large placental chorioangiomas are rare and may lead to pregnancy complications and poor perinatal outcomes, including fetal anaemia, hydrops fetalis, growth restriction, polyhydramnios and preterm delivery.[1] We report a case of a large placental chorioangioma, the antenatal management and the maternal and fetal outcomes.

Case Report

A 36-year-old gravida 5 healthy woman was diagnosed with a vascular placental tumour on ultrasound at 25 weeks of gestation. The tumour was 1 x 1 cm in size, with mixed echogenicity and well-defined margins. Until then, the patient had had an otherwise normal pregnancy with a negative serology for hepatitis and HIV and normal fetal anatomy scan. The woman had no significant past medical or surgical history. She had had two previous, full-term spontaneous vaginal deliveries and two spontaneous, first-trimester abortions. The initial scan evaluated in detail the placental function, characteristics of the tumour and the fetal anatomy. There were no fetal tumours or any other anomalies noted on that scan. Follow-up scans during pregnancy showed an increase in the tumour size, with it reaching a maximum of 6 x 5 cm at 32 weeks of gestation. The tumour was within the centre of the umbilical cord insertion with a central feeding vessel [Figures 1 & 2].

A provisional diagnosis of chorioangioma was made and the patient was assessed weekly in the high-risk clinic for any signs of maternal or fetal complications. The woman was well throughout the pregnancy with no antenatal complications such

Departments of [1]Obstetrics & Gynaecology and [2]Pathology, Sultan Qaboos University Hospital, Muscat, Oman
**Corresponding Author e-mail: drriyami@hotmail.com*

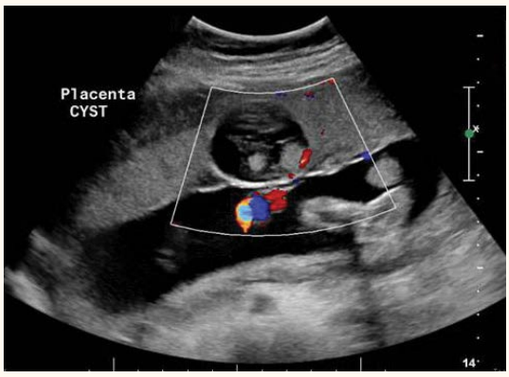

Figure 1: Placental chorioangioma of 6 x 5 cm in size within the centre of the umbilical cord with a feeding vessel.

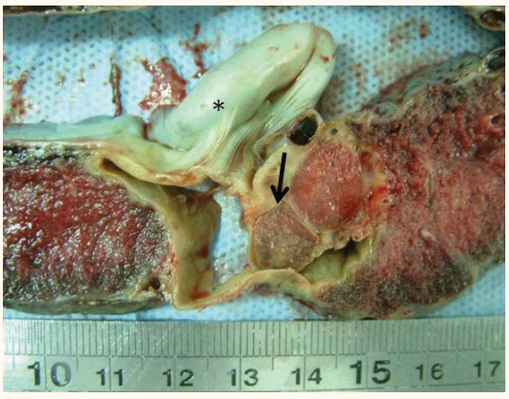

Figure 2: Well-circumscribed lesion (arrow) within the placenta just beneath the insertion of the umbilical cord (asterisk). It had a pale tan cut surface.

as pregnancy-induced hypertension or gestational diabetes. The fetus was monitored closely and the mother was also asked to monitor its movements. Growth scans were performed every two weeks to exclude intrauterine growth restriction (IUGR), and a weekly biophysical profile, done by Doppler, included measurements of the middle cerebral artery peak systolic velocity (MCA-PSV). The cardiac function of the fetus was assessed and, fortunately, there were no signs of compromise such as cardiomegaly, abnormal *ductus venosus* (DV) waveforms, or signs of fetal hydrops. There was no evidence of polyhydramnios, IUGR or fetal anaemia on follow-up scans. At 37 weeks and 3 days of gestation, the patient complained of reduced fetal movements; a scan showed a collapsed tumour 4 x 3 cm in size with less vascularity and irregular margins. There were no changes in the Doppler measurement of MCA-PSV or in cardiac function.

Labour was induced and the patient had a spontaneous vaginal delivery of a healthy male baby of 2,730 g with Apgar scores of 9 and 10 at 1 and 5 minutes, respectively. The postnatal assessment of the baby showed no evidence of anaemia or thrombocytopaenia. On macroscopic examination of the placenta, the umbilical cord was triple-vesselled and unremarkable, and the extraplacental membranes were translucent. The placental disc was 19 cm x 16 cm x 4 cm with a trimmed weight of 666 g. Beneath the insertion of the umbilical cord, a 2 cm pale-brown intraparenchymal lesion was noted adjacent to which was a cystic space; together, these measured 4.5 cm across.

Histopathological examination of the pale area showed an infracted chorangioma formed of a network of dilated cavernous type vessels lined by CD-31 positive endothelial cells with intervening collagenous stroma [Figure 3]. The cystic space did not have a true lining and its wall was continuous with the chorionic plate. Placental *villi* were appropriate for gestational age and were unremarkable.

Discussion

Placental chorioangiomas are the most common placental tumours, with a prevalence of 1%. They are thought to be hamartomatous lesions rather than true neoplasms and should be differentiated from other placental tumours.[1–3] Large placental chorioangiomas measuring more than 4 cm in size are rare, with an incidence of 1:500–1:16,000, and are associated with several perinatal complications including intrauterine fetal demise and premature birth.[1–8] The main differential diagnoses include chorangiosis, chorangiomatosis and rare chorangiomas with trophoblastic proliferation.

This case of a large placental chorioangioma was followed very closely during the antenatal period. Fortunately, no complications occurred for either the mother or the fetus, except that the chorioangioma became infarcted at 37 weeks of gestation, requiring induction of labour. It is known that large tumours may produce degenerative phenomena like necrosis, calcifications, hyalinisation and infarction. The infarction usually occurs spontaneously, leading to a decrease in echogenicity, tumour volume and blood flow on colour Doppler images, as observed in this case.[9]

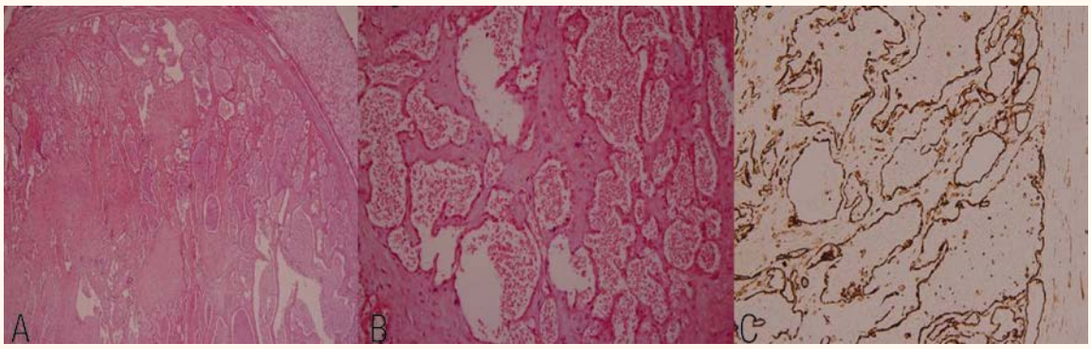

Figure 3 A to C: (**A & B**) Encapsulated lesion formed of variably-sized dilated and congested vessels. The lesion had undergone infarction. No chorionic *villi* were seen. (**C**) The CD-31 immunostain highlights the endothelial cells lining the vascular spaces.

The recurrence risk of chorangiomas is rare, but there are occasional reports of such cases, suggesting some environmental or genetic predisposition.[10] For example, there is a strong relationship between placental chorangiomas and gestation at high altitudes, suggesting the occurrence of vascular growth factors induced by hypoxia.[11]

Fetal growth restriction has been reported in association with large placental tumours.[12] The theory is not yet clear, but it could be due to these large vascular tumours acting as a physiological and functional dead space leading to placental insufficiency, chronic hypoxia, fetal compromise, fetal growth restriction and even death. Fetal anaemia could also result from these large tumours.[13] Close monitoring with Doppler measurement of MCA-PSV, and looking for early signs of hyperdynamic circulation, are essential actions as fetal cardiomegaly is important during the antenatal period. Early detection could prevent subsequent major complications, such as fetal death by antenatal interventions like *in utero* fetal blood transfusion and early delivery at a reasonable gestational age. Our patient was followed very closely with weekly scans looking for early signs of fetal anaemia and biweekly growth scans to rule out fetal growth restriction.

A large series of 19 cases with giant placental chorioangiomas was reported recently. Of those, 18 resulted in a variety of fetal complications, including fetal growth restriction in 6 cases and one with one stillbirth at 34 weeks' gestation due to severe placental insufficiency and the mother's refusal of an early delivery.[14] Polyhydramnios was reported in three cases, out of which two required amnioreduction.[14] This series also included the antenatal interventions performed in fetuses with evidence of hyperdynamic circulation to prevent the development of *hydrops fetalis*. The intervention included fetoscopic laser treatment in one case, preterm delivery in one case, and interstitial laser therapy in three cases.[14] Other possible interventions include fetoscopic devascularisation of the tumour by suture ligation of the arterial blood supply and alcohol ablation of the feeding vessel.[15,16]

Another group in Chile reported their experience with placental chorioangiomas, with 11 cases diagnosed over a 5-year period.[17] Polyhydramnios and preterm delivery were the most common complications. They concluded that amnioreduction in selected cases may improve the perinatal outcome; fetal hydrops carries the highest risk of perinatal death, and close follow-up of cases with no associated findings at the time of diagnosis is very important.

Conclusion

Large placental chorioangioma are rare and may lead to adverse perinatal outcomes. Prenatal diagnosis of these tumours with close follow-up during the antenatal period and early intervention is crucial, and may result in a healthy mother and fetus.

References

1. Amer HZ, Heller DS. Chorangioma and related vascular lesions of the placenta - a review. Fetal Pediatr Pathol 2010; 29:199–206.
2. Sepulveda W, Aviles G, Carstens E, Corral E, Perez N. Prenatal diagnosis of solid placental masses: The

value of color flow imaging. Ultrasound Obstet Gynecol 2000; 16:554–8.

3. Zalwl Y, Weisz B, Gamzu R, Schiff E, Shalmon B, Achiron R. Chorangiomas of the placenta: Sonographic and Doppler flow characteristics. J Ultrasound Med 2002; 21:909–13.
4. Taori K, Patil P, Attarde V, et al. Chorioangioma of placenta: Sonographic features. J Clin Ultrasound 2008; 36:113–15.
5. Wallenburg HCS. Chorioangioma of the placenta: 13 new cases and a review of literature from 1939 to 1970 with special reference to the clinical complications. Obstet Gynecol Surv 1971; 26:411–25.
6. Kurt B. Recent trends in chorangiomas, especially those of multiple and recurrent chorangiomas. Pediatr Dev Pathol 1999; 2:264–9.
7. Guschmann M, Henrich W, Entazami M, Dudenhausen JW. Chorioangioma - new insights into a well-known problem. 1. Results of a clinical and morphological study of 136 cases. J Perinatal Med 2003; 31:163–9.
8. Hamid A, Hadi JF, Strickland D. Placental chorioangioma: prenatal diagnosis and clinical significance. Am J Perinatol 1993; 10:146–9.
9. Reshetnikova OS, Burton GJ, Milovanov AP, Fokin EI. Increased incidence of placental chorioangioma in high-altitude pregnancies: Hypobaric hypoxia as a possible etiologic factor. Am J Obstet Gynecol 1996; 174:557–61.
10. Imdad A, Sheikh L, Malik A. A large chorioangioma causing intrauterine foetal demise. J Pak Med Assoc 2009; 59:580–1.
11. Kirkpatrick AD, Podberesky DJ, Gray AE, McDermott JH. Best cases from the AFIP: Placental chorioangioma. Radiographics 2007; 27:1187–90.
12. Mucitelli DR, Charles EZ, Kraus FT. Chorioangiomas of intermediate size and intrauterine growth restriction. Pathol Res Pract 1990; 186:455–8.
13. Haak MC, Oosterhof H, Mouw RJ, Oepkes D, Vandenbussche FP. Pathophysiology and treatment of fetal anemia due to placental chorioangioma. Ultrasound Obstet Gynecol 1999; 14:68–70.
14. Zanardini C, Papageorghiou A, Bhide A, Thilaganathan B. Giant placental chorioangioma: natural history and pregnancy outcome. Ultrasound Obstet Gynecol 2010; 35:332–6.
15. Quintero RA, Reich H, Romero R, Johnson MP, Gonzalves L, Evans MI. In utero endoscopic devascularization of a large chorioangioma. Ultrasound Obstet Gynecol 1996; 8:48–52.
16. Wanapirak C, Tongsong T, Sirichotiyakul S, Chanprapaph P. Alcoholization: The choice of intrauterine treatment for chorioangioma. J Obstet Gynecol 2002; 28:71–5.
17. Sepulveda W, Alcalde JL, Schnapp C, Bravo M. Perinatal outcome after prenatal diagnosis of placental chorioangioma. Obstet Gynecol 2003; 102:1028–33.