

INSIGHTS FROM A TEN-YEAR RETROSPECTIVE STUDY ON CHORANGIOMAS

Milka Kochoska¹, Blagica Krsteska¹, Daniela Bajdevska-Dukoska¹, Vladimir Risteski¹,
Gligor Ristovski¹, Daniel Milkovski², Slavica Kostadinova-Kunovska¹, Gordana Petrushevska¹

¹Institute of Pathology, Faculty of Medicine, University Ss. Cyril and Methodius in Skopje

²University Clinic of Gynecology and Obstetrics, University Ss. Cyril and Methodius in
Skopje

Abstract

The most common benign neoplasms of the placenta are non-trophoblastic tumors. The most common subtype is chorioangioma, but teratoma, leiomyoma, and hepatocellular adenoma are also seen. Giant chorioangioma is a rare tumor, with diameter larger than 4 cm and seen in association with elderly primipara, twin pregnancies, hypertension and diabetes. Giant chorioangioma is associated with complications that can affect both the mother and the fetus or neonate.

We present a retrospective study of chorangioma cases diagnosed at Institute of Pathology in Skopje in a 10-year period from 2012 to 2021.

Macroscopic observations, gestational age at delivery, maternal age, and pregnancy outcomes were assessed. Histological and immunohistochemical examinations of the placental chorangiomas were conducted. Six cases of chorangiomas were clinically identified and histopathologically confirmed. Four cases involved giant chorangiomas ranging in size from 8 to 13 cm. One case was identified as chorangioma of the umbilical cord. Three of the cases showed potential maternal risk factors like primary infertility, extreme obesity and post COVID status. Four of the pregnancies had favorable outcomes with no complications during and after birth, one of the pregnancies resulted in a premature birth, while the other was complicated by fetal distress.

Regular monitoring by serial ultrasound and fetal echocardiography is recommended to pick up complications early so that they can be dealt effectively.

Key words: chorangioma, placental tumors, pregnancy outcome

Introduction

The most common benign neoplasms of the placenta are non-trophoblastic tumors such as chorioangioma, teratoma, leiomyoma, and hepatocellular adenoma. Chorioangioma is the most common subtype [1]. The incidence of chorangioma is 0.5–1.0% [2]. Small chorangiomas are clinically insignificant [3], while giant chorioangiomas are rare tumors, measuring more than 4 cm in diameter. They are often found in elderly primipara, twin pregnancy, hypertension and diabetes, and may be associated with complications that can affect both the mother and the fetus or neonate [4,5].

Case series

We present a series of chorangioma cases for which size of the lesion, histopathological characteristics, gestational weeks at delivery, maternal age and outcome of pregnancy were evaluated. Six cases of placental chorangiomas were diagnosed at Institute of Pathology, Faculty of Medicine in Skopje during a period of 10 years from 2012 to 2021. Four cases were giant chorangiomas with diameter from 8 to 13 cm. One of the cases was identified as chorangioma of the umbilical cord. (Table 1)

Three of the cases showed potential maternal risk factors like primary infertility, extreme obesity and post COVID status.

The gestational age of delivery ranged between 33 weeks and 6 days (33+6 weeks) and 39 weeks and 6 days (39+6 weeks). The maternal age range was between 25 to 34. Four of the pregnancies had favorable outcome with no complications during and after birth. One of the pregnancies resulted in a premature birth, while the other was complicated by fetal distress. (Table2)

Table 1. Placental chorangiomas in ten-year period (2012-2021)

Year	Gross specimen	Tumor measurements	Tumor location	Risk factors	Outcome
2012	Placenta	1,2 x 1,2 cm	Umbilical cord	Undetermined	No complications
2013	Placenta	8 x 7 x 6 cm	Subamniotic, above the surrounding level of the placental plate	Undetermined	No complications
2014	Tumor formation	9 x 8 x 5 cm	Undetermined	Undetermined	No complications
2016	Placenta	13 cm	Subamniotic and Intraplacental	St. post infertilitas primaria, St. post IVF et ET	Foetal distress
2021	Placenta	2,5 x 1 x 1 cm	Undetermined	Extreme obesity, St. post COVID	No complications
2021	Placenta	8 x 8 x 7 cm	Subamniotic, next to the umbilical cord	Prematurity	Premature delivery

Table 2. Clinical features

Year	Gestational weeks	Gender	APGAR score	Neonatal weight and lenght	Maternal age	Delivery
2013	ml X (39,6 g.n.)	female	9/10	3260 gr 51cm	26	Spontaneous
2014	ml X (39 g.n.)	female	8/9	3150 gr	34	Spontaneous
2016	ml X (39 g.n.)	male	8/9	3050 gr 50 cm	31	Cesarean section
2021	ml X (37+4 g.n.)	male	8/9	3940 gr 51 cm	26	Cesarean section

2021	ml IX (33+6 g.n.)	female	7/7	2170 gr 45 cm	25	Spontaneous
------	----------------------	--------	-----	------------------	----	-------------

Gross findings:

Macroscopically, most of the cases showed well-circumscribed nodular lesions with a red-brown coloration and a fleshy appearance, measuring from 1.5 cm to 9 cm in diameter. (Figure 1)



Figure 1. A giant chorangioma measuring 8 cm in diameter.

Microscopic findings:

Microscopic examination of the mass showed numerous proliferated thin walled capillaries lined by flattened endothelium and separated by fibrous stroma (Figure 2). This was further confirmed by immunohistochemistry for CD34, which showed strong reactivity of endothelial cells (Figure 3).

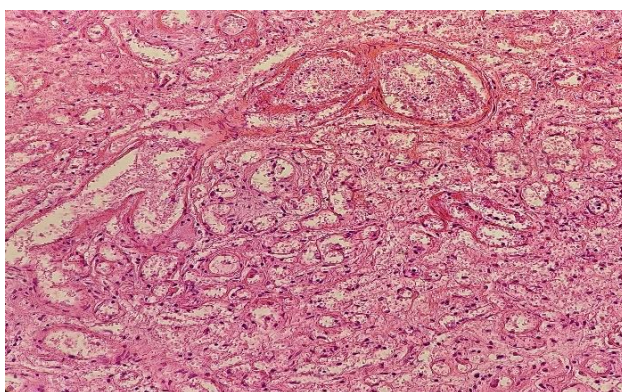


Figure 2. HeEoX200

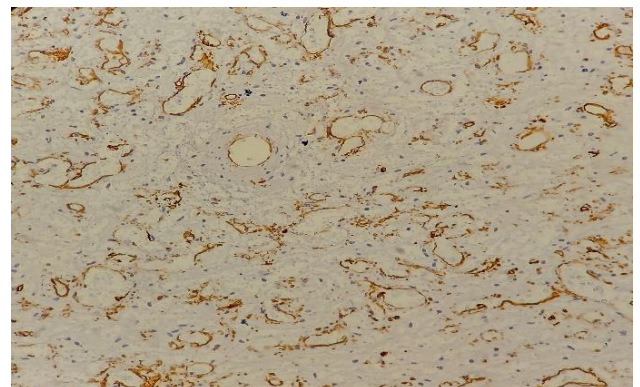


Figure 3. CD34X200

Discussion

Chorangioma is a benign non-trophoblastic neoplasm of the placenta, often seen as solitary nodules, mostly bulging from the fetal surface of the placenta or attached to the placental disc by a pedicle with a greater frequency in less perfused areas such as chorionic plate and placental margins [6].

Chorangiomas tend to arise in hypoperfusion areas. This is believed to result from a hypoxic condition driven by vascular growth factors, leading to active connective tissue proliferation and the expansion of villous capillaries. Pre-eclampsia, high-altitude pregnancy, and fetal anemia were also associated with the occurrence of chorangioma, suggesting that decreased oxygen tension may play an important role in the pathogenesis [7].

Microscopically, three histological patterns are recognized: angiomatous, cellular, and degenerate. The angiomatous pattern, being the most common, consists of numerous proliferating blood vessels at different stages of differentiation, ranging from capillary to cavernous, within placental stroma. Immunohistochemically, the tumour cells are positive for CD31, CD34, factor VIII, GLUT1 and cytokeratin 18, findings that suggest origin from blood vessels of the chorionic plate and anchoring villi.

The differential diagnosis of chorangioma includes chorangiosis and chorangiomatosis, which present as diffuse or, more commonly, focal proliferation of villous angioblastema, with villi absent in chorangioma. Another, less common differential is chorangioma with trophoblast proliferation (referred to as "chorangiocarcinoma", though likely a misnomer), characterized by a rare proliferation of trophoblastic tissue near an otherwise benign chorangioma [8]. These lesions are occasionally categorized as placental hamartomas rather than true neoplasms and have no malignant potential [9].

Diagnosis is primarily performed using ultrasonography, the main diagnostic method, which reveals a solid mass within the placenta characterized by irregular echogenicity [10]. Through color Doppler, it is possible to observe both feeding vessels that enter the placental mass and diffuse peritumoral vascularization [11].

The management of chorioangioma is typically conservative. While no specific treatment is required for asymptomatic cases, careful monitoring through serial ultrasound examinations is essential to detect early complications [12].

Pregnancies with small placental chorioangiomas are often asymptomatic and typically do not affect the course of pregnancy. In contrast, large placental chorioangiomas are more likely to be associated with perinatal complications and adverse fetal outcomes. These may include polyhydramnios, preterm labor, fetal anemia, fetal growth restriction, fetal cardiomegaly or heart failure, fetal hydrops, maternal mirror syndrome, and, in severe cases, fetal demise [13].

Because of severe fetal sequelae and poor perinatal outcome with large tumors, treatment may include an attempt to reduce blood flow to the tumor by means of vessel occlusion or ablation [14].

Conclusion

Placental chorangioma is an uncommon tumor that presents challenges due to its potential for serious complications, which may adversely affect pregnancy outcomes. Reporting cases of placental chorioangioma is crucial to enhance understanding and help estimate the actual rates of maternal-fetal morbidity and mortality. Additionally, thorough examination of the placentas, including detailed macroscopic and microscopic analysis by the pathologist, is essential to provide a more accurate explanation to family members and healthcare professionals about the potential causes of pregnancy loss or complications.

References:

1. Zacharis K, Kravvaritis S, Charitos T, Chrysafopoulou E, Fouka A. A rare case of a giant placental chorioangioma with favorable outcome. *Pan Afr Med J.* 2020;36:214. doi:10.11604/pamj.2020.36.214.23630.
2. Wallenburg HC. Chorangioma of the placenta: thirteen new cases and a review of the literature from 1939 to 1970 with special reference to clinical complications. *Obstet and Gynecol Survey.* 1971;26:411–25. doi: 10.1097/00006254-197106000-00001.
3. Benirschke K, Burton GJ, Baergen RN. *Pathology of the Human Placenta.* 6th ed. Springer; 2012. p. 747-754.

4. Patil RN, Helwatkar S, Raut W, Gadodiya K. Giant chorioangioma: A case report. *J Med Sci Health*. 2017;3(3):22-25. doi:10.46347/jmsh.2017.v03i03.003.
5. Moreno-Lopez JC, Gutierrez-Sanchez EF, Herrera-Barrera LE, Murillo-Bargas H. Giant placental chorioangioma in early term pregnancy: A case report. *Ginecol Obstet Mex*. 2021;89(8):635-640. doi:10.24245/gom.v89i8.5025.
6. Akbarzadeh-Jahromi M, Soleimani N, Mohammadzadeh S. Multiple chorangioma following long-term secondary infertility: A rare case report and review of pathologic differential diagnosis. *Int Med Case Rep J*. 2019;12:383-387. doi:10.2147/IMCRJ.S222462.
7. Theresia E, Nurdianti D, Widodo I. Giant placental chorangioma: The first case report in Indonesia. *Hum Pathol Case Rep*. 2021;23(1):200472. doi:10.1016/j.ehpc.2021.200472.
8. Kataria N, Singh A, Bedi PK. Giant placental chorangioma: A rare case report. *J Clin Diagn Res*. 2016 Apr;10(4):ED03-4. doi:10.7860/JCDR/2016/17222.7540.
9. Kodandapani S, Shreshta A, Ramkumar V, Rao L. Chorioangioma of placenta: a rare placental cause for adverse fetal outcome. *Case Rep Obstet Gynecol*. 2012;2012:913878. doi:10.1155/2012/913878.
10. Torres-Correa JE, Sánchez-Montoya MA, Sandoval-Sánchez J, Castro-Álvarez JF. Corioangioma placentario: reporte de un caso en la Unidad de Patología del Hospital San Juan de Dios E.S.E. Rionegro-Antioquia. *Med Lab*. 2020 Sep;24(4):325-332. doi:10.36384/01232576.340.
11. Bastonero S, Sciarrone A, Galtarossa G, Pertusio A, Dusini I, Botta G, Benedetto C. Ultrasound features of placental chorioangioma detected by SMI technology before and after thrombosis of feeding vessels: analysis of a clinical case. *Perinatal Journal*. 2022;30(2):200-205. doi:10.2399/prn.22.0302007.
12. Abdalla N, Bachanek M, Trojanowski S, Cendrowski K, Sawicki W. Placental tumor (chorioangioma) as a cause of polyhydramnios: a case report. *Int J Womens Health*. 2014 Nov 20;6:955-9. doi: 10.2147/IJWH.S72178.
13. Ma H, Liu Z, Ruan J. Placental chorioangioma and pregnancy outcome: a ten-year retrospective study in a tertiary referral centre. *BMC Pregnancy Childbirth*. 2023 May 25;23(1):381. doi: 10.1186/s12884-023-05719-x.
14. Panat V. Chorioangioma: a case report. *J Fetal Med*. 2020 Dec;7(4):325-329. doi:10.1007/s40556-020-00270-7.