



## Five too many! A Case of Incidentally Detected Five Vessel Umbilical Cord Associated with Placental Chorangiomas

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### Abstract

Supernumerary vessels are defined as presence of four or more vessels present within the umbilical cord. This condition may often accompany a poor pregnancy outcome and presence of congenital anomalies. They may be present in healthy fetuses. Chorangiomas are an adaptive response to in-utero hypoxia and its presence signifies better pregnancy outcomes. Abnormalities that lead to multiple cord vessels are rare with the majority of reported cases highlighting four vessels due to a persistent right umbilical vein. Here, we report a case of a 5-vessel umbilical cord associated with chorangiomas of placenta, detected incidentally after birth.

**Keywords:** Blood vessels, Umbilical cord, placenta, placenta diseases

### Introduction

The umbilical cord (UC) is an important vascular organ maintaining fetal well-being and development. [1] Abnormalities of the UC, referring to its morphology, placental insertion, number of vessels and primary tumors, may have a direct impact on the perinatal outcome and may relate with fetal abnormalities. [2] Chorangiomas are an uncommon pathology seen in 5-6% of placentas, with its incidence increasing with the gestational age. It is known to occur as an adaptive response to chronic placental hypoxia and has been found to be associated with various maternal, fetal and placental disorders. Rarely, its association has been demonstrated with UC abnormalities such as true and false knots, long cord, umbilical vein dilatation or thrombosis, nuchal cord, and single umbilical artery. [3] Here, we report a case of chorangiomas of placenta associated with the a rare 5-vessel umbilical cord.

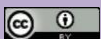
### Case Report

A 32-year-old primigravida, presented to the Gynecology OPD in March 2022, with chief complaint of discharge of significant amount colorless fluid from her vagina since two days. Obstetric examination revealed a Premature Rupture of Membranes (PROM), estimated to have occurred for

more than 48 hours. On further evaluation, her gestation period was found to be at 39 weeks, and she did not present with any other co-morbidities associated with pregnancy. She was an unregistered case, and hence did not have any previous ultrasonography reports for reference. She was immediately admitted and taken for elective LSCS (Lower Segment Caesarean Section) given of prolonged PROM.

The surgery concluded with the delivery of a healthy neonate. It cried immediately at birth, and neonatal evaluation revealed no congenital disability or feature suggestive of aneuploidy or other genetic defects. However, because of prolonged PROM, the baby was administered intravenous antibiotics prophylactically, while the placenta was sent for histopathological evaluation to rule out chorioamnionitis.

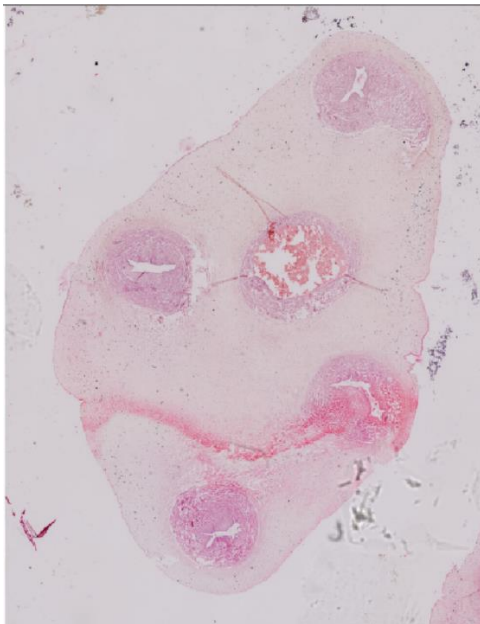
The department of histopathology of the department of laboratory sciences received a placenta with attached UC, with the placental dimensions being 13x10x3cm and the UC measuring 29 cm. On sectioning of the UC, 5 umbilical vessels were noted on gross examination (Figure 1). The placenta did not show any gross abnormalities, and its membranes could be easily stripped off. No thickening of the membranes, any purulent deposits, or retroplacental clots were noted.



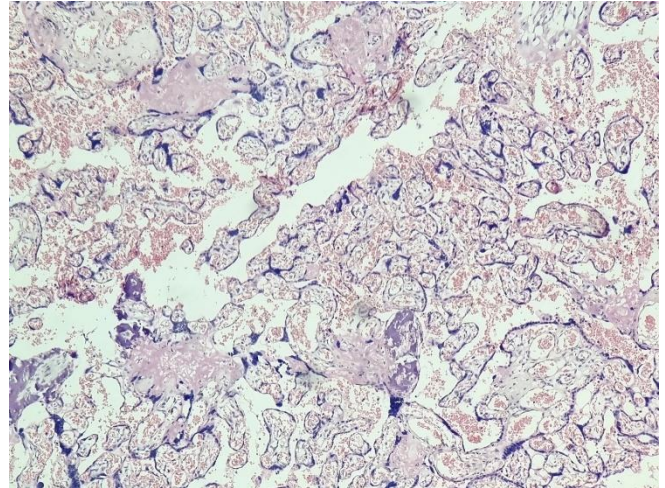
Microscopic evaluation of the UC revealed the presence of supernumerary vessels (Figure 2). These vessels included four umbilical arteries and an umbilical vein. The membranes had minimal inflammatory infiltration, ruling out possible chorioamnionitis. The placental cotyledons; however, showed increased vascularity associated with increased syncytial trophoblasts and a few calcific foci, secondary to hemorrhage (Figure 3 and 4). Further placental cotyledons evaluation revealed the presence of more than ten capillaries in more than ten terminal villi in ten different non-infarcted areas in at least three low power fields. These features connoted a diagnosis of placental chorangiosis.



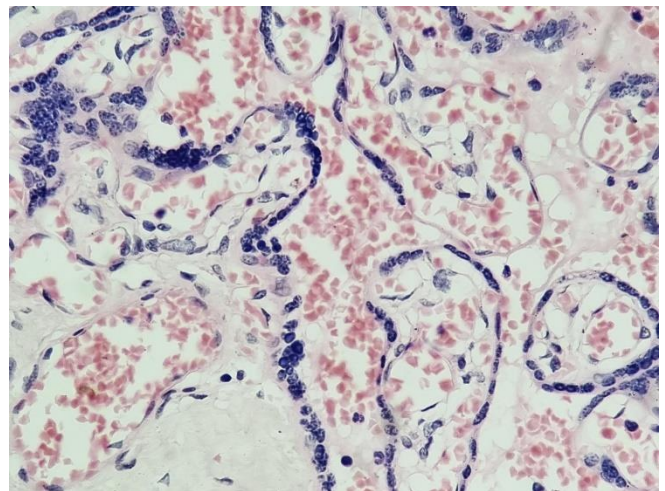
**Figure 1:** Gross cut section of the umbilical cord, with the cut surface showing five vessels;



**Figure 2:** Photomicrography of the umbilical cord showing five umbilical vessels (H&E; 10x)



**Figure 3:** Photomicrography of the placenta: - cotyledons showing a few calcific foci in the hemorrhagic areas along with presence of increased villous vascularity (>10 vessels present in >10 villi in non-infarcted areas) associated with increased syncytial knots



**Figure 4:** Photomicrography of the placenta: High power view of the chorangiotic placental cotyledons (H&E; 400x)

The overall histopathology of the placenta concluded with a five-vessel umbilical cord with chorangiosis.

The neonate remained stable and continued to breastfeed normally. No features suggestive of any congenital anomaly or a genetic disability were noted during the hospital stay. The mother and neonate were discharged and scheduled on a regular follow-up.

## Discussion

The placenta is an important materno-fetal connection that is responsible for nourishing and protecting the fetus during

Table 1: Cases of five vessel UC reported in literature till date

Author	Year	Country	No of cases	Period of gestation (weeks)	Type of pregnancy	No of vessels in the UC	Type of vessels	Associated abnormalities	Outcome
Cohen et al <sup>10</sup>	1992	United States of America (USA)	1	32	Twin pregnancy	5	3 arteries, 2 veins	Thoracopagus conjoined twins with conjoined hearts and shared liver	Both the babies succumbed during surgical separation of the heart and liver.
Singh et al <sup>1</sup>	2012	India	3	1. 33 2. 36 3. 16	1. Diamniotic monochorionic twins 2. Diamniotic dichorionic twins 3. Singleton	a. 1 <sup>st</sup> twin: 3 2 <sup>nd</sup> twin: 5 b. 1 <sup>st</sup> twin: 3 2 <sup>nd</sup> twin: 5 c. 5	a. 1 <sup>st</sup> twin: NR 2 <sup>nd</sup> twin: NR 4 arteries, 1 vein  b. 1 <sup>st</sup> twin: NR 2 <sup>nd</sup> twin: 3 arteries, 2 veins  c. 2 arteries, 3 veins	a. 1 <sup>st</sup> twin: NR 2 <sup>nd</sup> twin: None  b. 1 <sup>st</sup> twin: NR 2 <sup>nd</sup> twin: None  c. Anencephaly with open spina bifida	NR  NR  NR
Nallasivam et al <sup>11</sup>	2016	India	1	NR	Singleton	5	4 arteries, 1 vein	None	NR
Paramanatham et al <sup>12</sup>	2018	India	3	NR	1. Singleton 2. Twin pregnancy 2. Twin pregnancy	a. 5 b. 5 c. 5	NR	a. Anencephaly b. None c. None	NR AW AW
Garg et al <sup>13</sup>	2018	India	1	34	Singleton	5	4 arteries, 1 vein	Chorangiosis of placenta with presence of omphalomesenteric duct remnant	Stable at birth
This case	2022	India	1	32	Singleton	5	4 arteries, 1 vein	Chorangiosis of placenta	AW
<b>Total cases</b>	10								
<b>AW = Alive and well; NR = Not reported</b>									

pregnancy. Its histopathological examination can reveal significant information about pre-uterine and uterine conditions affecting fetal growth during pregnancy [3]. The UC is flexible cord like structure responsible for maintaining fetal blood circulation during pregnancy. The prenatal well as the postnatal examination of the placentas contributed significantly to the current knowledge of UC anomalies and their impact on fetal outcome. Hence, the examination of the number of vessels in the UC has become a standard part of routine pre-natal ultrasonography as well as gross examination of the placenta [2].

The UC normally contains three vessels: two umbilical arteries (UA) and one umbilical vein (UV) [2]. The presence of more than three vessels, apart from those mentioned above is defined as supernumerary umbilical vessels [4]. The incidence of supernumerary vessels is 1%; of these, most are four vessel UCs with the presence of a persistent Right

Umbilical Vein (PRUV). Presence of an additional umbilical artery is rare. Similarly, literature mentions very few cases of a five-vessel UC reported so far.

Our case presents with the presence of four UAs and one UV. The additional umbilical vessels are thought to arise from persistent vitelline vessels [5].

Supernumerary umbilical artery is extremely rare. Only occasional reports mention the presence of five blood vessels in the UC. The association of five-vessel cord with congenital disorders is not clearly comprehended because of the limited literature present pertaining to this condition. The presence of five or more vessels in the UC has usually been described as related to conjoined twins. Consequently, this condition does not always herald an adverse perinatal outcome. However, this rarely reported vascular pathology may not always be associated with congenital

malformations, but such cases require a comprehensive antenatal workup, to permit a better perinatal outcome. Unfortunately, our case did not have any previous work-up records available with her since she was an unregistered case. Table 1 describes the cases of five vessel UCs reported in literature so far.

Chorangiosis is defined as capillary hyperplasia in terminal villi due to chronic placental hypoperfusion or low-grade tissue hypoxia. It is essentially terminal villous vascular hyperplasia resulting from longstanding low-grade hypoxia in the placental tissue or fetal side hypoperfusion [6].

It is commonly associated with women living in high altitudes, maternal anaemia and smoking in pregnancy. It may also be seen in pregnancies complicated by pre-eclampsia, diabetes mellitus, certain infections, multiple gestations, cardiovascular and respiratory diseases, air pollution and obesity [6].

The criteria for diagnosis of chorangiosis was given by Altshuler [7]. It denotes the presence of > 10 capillaries in at least 10 terminal villi in  $\geq 10$  non-infarcted areas in at least 3 low power fields of the placenta. Normally, the villi rarely have more than five capillaries per villous.

The presence of chorangiosis is associated with an increased fetal morbidity and mortality and may commonly present with low Apgar scores, fetal neurocompromise, fetal growth restriction, congenital malformations or even neonatal death [6,8].

Literature mentions a few theories for the pathogenesis of chorangiosis. One of them proposes that chronic hypoperfusion or tissue hypoxaemia leads to elaboration of vascular endothelial growth factor, platelet-derived growth factor, and transforming growth factor-beta by mesenchymal and trophoblastic cells; while the other hypothesis highlights the role of macrophage-derived tumor necrosis factor- $\alpha$ . Increased intramural pressure due to umbilical vein obstruction is also thought to play a role in the development of chorangiosis in cases associated with cord anomalies, such as long umbilical cord and thrombosis of vessels [3,9]

Very sparse literature is available regarding association of supernumerary vessels of UC with chorangiosis. Garg et al [3] have presented a case of a five vessel UC associated with chorangiosis, where the mother presented with gestational hypertension with associated oligohydramnios; however,

they could not establish a definite correlation as well. Among the other cases of five vessel UCs reported in literature, only Cohen et al [10] have mentioned about a twin gestation associated with Siamese twins, where the mother reported to the hospital with a history of large for gestation abdominal girth. Other authors do not mention about the maternal history, although a definite mention is made regarding the presence of an associated birth defect in the new-born with a five vessel UC. Compared to the cases in literature where the maternal history has been mentioned, this case did not present with any maternal co-morbidities which might associate with the development of a multivessel UC. This case, is the second case to be reported in English literature, where a five vessel UC is associated with placental chorangiosis, after Garg et al in 2018.

## Conclusion

Supernumerary umbilical vessels always call for a detailed examination of the placenta as well as a complete maternal and fetal clinical evaluation to rule out the presence of gross as well as genetic abnormalities. Chorangiosis is an important marker of intrauterine fetal hypoxia, and although its presence signifies a better pregnancy outcome, the presence of chorangiosis does herald a complete neonatal examination and evaluation to rule out the presence of hypoxia associated injuries. More cases of supernumerary UC with placental chorangiosis need to be studied and reported to have a better understanding of the association between supernumerary vessels and placental chorangiosis.

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## Reference

1. Singh N, Rao S, Sobti P, Khurana N. Multiple vessels in the umbilical cord: A report of four cases. *Indian Journal of Pathology and Microbiology*. 2012;55(4):597–8.
2. Damiani GR, del Boca G, Biffi A. Five-vessel umbilical cord and fetal outcome: an obstetric overview. *Journal of Maternal-Fetal and Neonatal*

- Medicine. 2021;
3. Garg N, Diwaker P, Aggarwal S, Gaur JH. Chorangiosis placenta with 5-vessel umbilical cord with omphalomesenteric duct remnant: An unusual association. *Turkish Journal of Obstetrics and Gynecology*. 2018;15(4):270–2.
  4. Senyuva I, Küçük S. A partial supernumerary umbilical vessel. *Journal of Clinical Obstetrics and Gynecology*. 2020 Aug 11;30(2):78–81.
  5. Ziadie MS. Placenta Nonneoplastic placental conditions and abnormalities Umbilical cord Single umbilical artery and supernumerary vessels. *PathologyOutlines.com*. 2020. p. 1–2.
  6. Huynh A, Roberts D. Placenta: Nonneoplastic placental conditions and abnormalities - Noninfectious - Chorangiosis. *PathologyOutlines.com*. 2021. p. 1–4.
  7. Altshuler G. Chorangiosis. An important placental sign of neonatal morbidity and mortality. *Arch Pathol Lab Med*. 1984 Jan;108(1):71–4.
  8. Barut A, Barut F, Kandemir NO, Aktunc E, Arikan I, Harma M, et al. Placental chorangiosis: The association with oxidative stress and angiogenesis. *Gynecologic and Obstetric Investigation*. 2012 Mar;73(2):141–51.
  9. DoğanGün B, Barut F, AlperTanriverdi H, Özdamar SO, Barut A. Placental Chorangiosis: An Important Pattern of Placental Injury. *Gynecology Obstetrics & Reproductive Medicine*. 2006;12:176–9.
  10. Cohen HL, Shapiro ML, Schwartz D. The Multivessel Umbilical Cord: An Antenatal Indicator of Possible Conjoined Twinning. Vol. 20, *J Clin Ultrasound*. 1992.
  11. Nallasivam D, Kuruvila SK. A Study Of Correlation Between Placental And Umbilical Cord Abnormalities And Foetal Outcome Of Patients Delivering At A Tertiary Care Hospital. *Journal of Evidence Based Medicine and Healthcare*. 2016 Jul 4;3(53):2738–40.
  12. Paramanatham P, Swarna S. Multivessel Umbilical Cord: Case Report. *Mod Appl Bioequival Availab [Internet]*. 2018;3(1):001–2.

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