Cureus

Open Access Case Report

DOI: 10.7759/cureus.51273

Double-to-Single Umbilical Artery With a Low Pulsatility Index Leading to Fetal Death: A Case Report and Review of Literature

Review began 12/12/2023
Review ended 12/23/2023
Published 12/29/2023

© Copyright 2023
Lin et al. This is an open access article distributed under the terms of the Creative Commons Attribution License CC-BY 4.0., which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Yanting Lin [1], Ruiling Yan [1], Andong He [1], Jie Chen [2], Ruiman Li [1]

1. Obstetrics and Gynecology, The First Affiliated Hospital of Jinan University, Guangzhou, CHN 2. Obstetrics and Gynecology, The Affiliated Shunde Hospital of Jinan University, Guangzhou, CHN

Corresponding author: Ruiman Li, hqyyck@126.com

Abstract

Single umbilical artery (SUA) may be associated with adverse pregnancy outcomes, such as fetal death, emergency cesarean section, premature delivery, small-for-gestational-age infants, and admission to neonatal intensive care unit, and some SUAs are transformed from originally double umbilical arteries (UA). The pulsatility index (PI) can reflect the resistance of UA, and clinicians attach importance to high PI but easily overlook low levels of it. We reported one case of a pregnant woman who underwent double to single UA accompanied by low UA-PI and finally had intrauterine fetal death. Additionally, the literature regarding SUA and UA-PI is reviewed. This study aims to alert clinicians to the risk of double-to-single UA with low UA-PI and strengthen fetal monitoring and timely intervention. We look forward to more clinical evidence to investigate it.

Categories: Obstetrics/Gynecology
Keywords: intrauterine fetal death, umbilical artery, adverse outcome, pulsatility index, single umbilical artery

Introduction

There are two umbilical arteries and one umbilical vein in a normal umbilical cord; when there is only one umbilical artery (UA), it is called a single umbilical artery (SUA), which is the most common cord malformation with an incidence of approximately 5.3%-4.0% [1]. Its occurrence may be associated with various factors, including the proliferation, necrosis, and thrombosis of vascular smooth muscle cells, the hypercoagulable state of pregnant women, and umbilical cord torsion [2]. The SUA is prone to increased pulsation index (PI), which attracts the attention of clinicians; a low PI of SUA is easily overlooked because it is believed to mean a low blood flow resistance, although it is sometimes a sign that blood flow is close to interruption. In this study, we present one clinical case in which her fetus had double umbilical arteries in the beginning, but the umbilical arteries became SUA with low PI in the third trimester and finally had an intrauterine stillbirth.

Case Presentation

In November 2021, SUA (the left side) (Figure *1*) accompanied by low UA-PI at 0.67 (1.5th) and UA-resistance index (RI) at 0.48 (0.7th) was noted during regular antenatal care at 30+6 weeks of gestation in a 33-year-old multigravida (Figure *2*).

How to cite this article
Lin Y, Yan R, He A, et al. (December 29, 2023) Double-to-Single Umbilical Artery With a Low Pulsatility Index Leading to Fetal Death: A Case Report and Review of Literature. Cureus 15(12): e51273. DOI 10.7759/cureus.51273

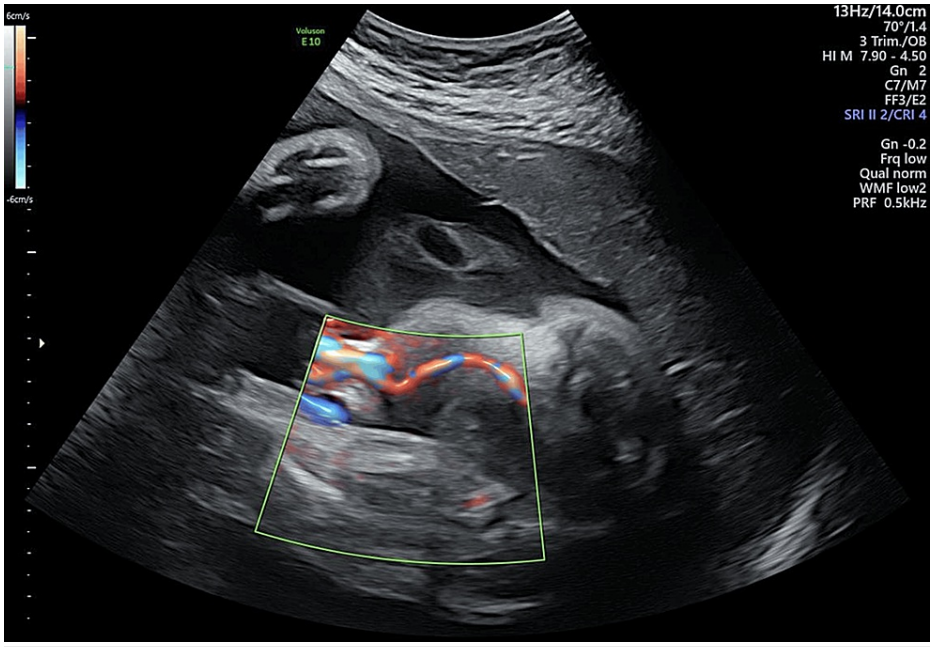


FIGURE 1: Single umbilical artery at 30+6 weeks (using a color Doppler ultrasound diagnostic system; Voluson™ E10, GE HealthCare, USA)



FIGURE 2: The red and blue dots indicate the corresponding umbilical artery pulsation index at that time point

Low UA-PI, i.e., UA-PI < 2th percentile for gestational age. We use a WeChat mini program Yunsuan to obtain the percentile of the UA-PI and UA-resistance index (PI) for the corresponding gestational weeks. The woman's last pregnancy ended up with a missed abortion for an unknown reason at 13 weeks. Her progestational body mass index (BMI) was 25.32 kg/m^2, showing she was overweight. There were no other structural abnormalities detected. Nuchal translucency was 2.9 mm at 13+3 weeks. Other routine investigations during the first and second trimesters were normal, and there were still double umbilical arteries at 20+6 weeks (Figure *3*).

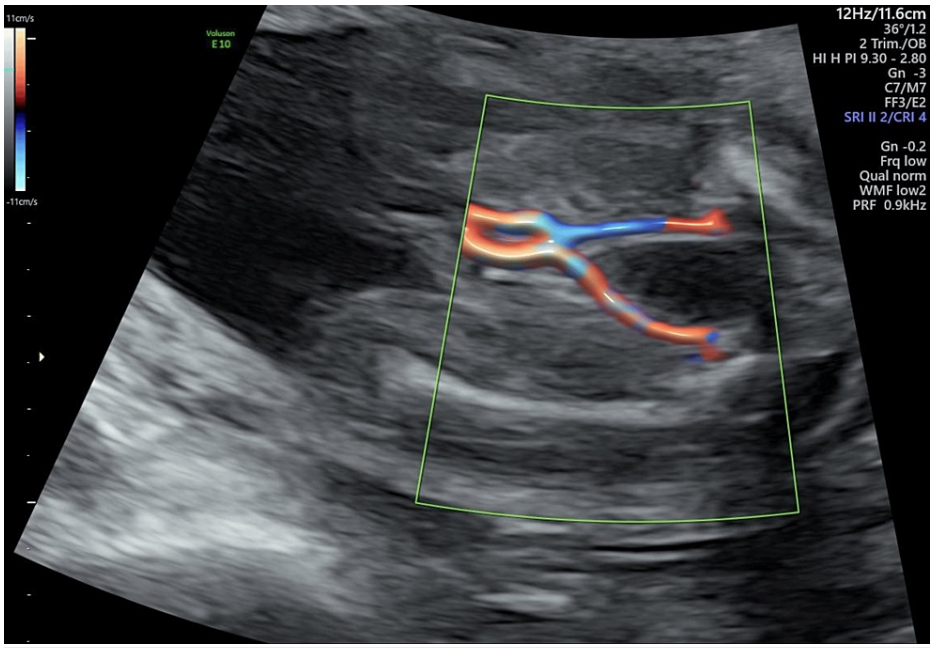


FIGURE 3: Double umbilical arteries at 20+6 weeks

Due to the thick nuchal translucency, fetal two-dimensional echocardiography was done at 20+6 weeks of gestation to rule out any associated cardiac anomalies, and a four-dimensional color Doppler ultrasound was done at the same time routinely. The report was normal and no cardiovascular, urinary, or reproductive system malformations or other abnormalities were detected. The woman was operated on for amniocentesis at 31 weeks on account of the SUA found at 30+6 weeks. Because no chromosome abnormalities were found in the amniocentesis and the fear of contracting novel coronavirus, she missed her regular appointment at 33 weeks. At 34+5 weeks, she felt more than half of the fetal movement decreased, and the next day she came to our department, the hemodynamic parameters showed fetal death. The 2.10 kg stillborn female fetus' appearance was normal. The 33 cm umbilical cord was twisted around the neck for one circle. The pathological examination showed cord edema. The patient was informed, and consent was taken before submitting this case report. Table *1* lists the details of each antenatal visit.

Date	Gestational age (weeks)	Maternal weight (kg)	Fundal height (cm)	Maternal abdominal circumference (cm)	Fetal heart rate (bpm)	Estimated fetal weight (g)	Other findings
11/4	13+3	64.6	-	-	166	75	NT 2.9 mm
11/5	17+5	-	-	-	visible	-	The blood examinations were normal
2/6	20+6	-	-	-	157	342	Fetal echocardiography and four-dimensional Doppler ultrasound were normal
14/7	26+6	67.5	-	-	visible	-	-
4/8	29+6	68.5	27	96	150	-	-
12/8	31	69.5	28	96	145	1553	The result of the amniocentesis was normal
8/9	34+6	72	31	100	0	2293	Fetal death

TABLE 1: Detailed information at each antenatal visit

Discussion

Currently, there are two theories for the occurrence of SUA: one is that it is congenitally underdeveloped, and there is only one UA from the beginning of embryonic development [3]; the other is that there are two umbilical arteries at the beginning of embryonic development, but one of the umbilical arteries atrophies and disappears secondarily in the later development process. The potential reasons for the change of umbilical vessels from double umbilical arteries to SUA are as follows: (1) thrombosis and embolism: thrombus can form in umbilical vein or artery circulation. The etiology of umbilical vessel thrombosis can be explained by Virchow's triad of reduced blood flow velocity, hypercoagulable state, and vascular abnormalities. The decrease in blood flow velocity may be related to mechanical damage of the umbilical cord or abnormally anatomical structure of the umbilical cord such as umbilical cord torsion, slim and narrow cord, umbilical cord entanglement, umbilical cord overlength, umbilical cord true knot, umbilical cord compression [4]. In this study, an umbilical cord twist was found in this case. Studies have shown that low UA-PI in SUA, abnormal cord blood flow, and thrombosis are mutually reinforcing, leading to a vicious cycle and ultimately vascular occlusion [5]. (2) Immune system disorder [6]: the anti-SSB/La antibody, rheumatoid factor, antinuclear and lack of protein S may be related to immune system diseases, such as primary Sjogren's syndrome, systemic lupus erythematosus, congenital heart disease, and neonatal lupus, which are high-risk factors for thrombosis. (3) Release of inflammatory factors: inflammatory factors can lead to vascular endothelial damage, which promotes thrombosis [7]. There are other risk factors related to SUA, such as gestational hypertension, history of diabetes, smoking history, or obesity [3]. In this case, the woman's progestational body mass index (BMI) was 25.32 kg/m^2, showing she was overweight. Due to the low incidence of double UA becoming SUA in the third trimester and the limited research data, there is no consensus on prenatal monitoring and treatment strategies. Notably, UA-PI reduction is often overlooked because the UA-PI will decrease with the increase of gestational weeks in the normal process of pregnancy, but the flow rate of UA can be extremely low when the blood flow is close to cut-off. This case resulted in intrauterine fetal death due to untimely treatment. Prenatal fetal monitoring can reduce the risk of stillbirth, including fetal movement monitoring and fetal ultrasound Doppler examination. Decreased fetal movement was found in this case. Besides, although no chromosome abnormalities were found in the amniocentesis, regular antenatal care is necessary. Therefore, this study aims to provide cautionary evidence about the clinical management of SUA, particularly SUA with low PI, to raise the profile of clinicians on it.

Conclusions

In summary, the timely detection of UAs and their blood flow is of great importance to ensure fetal safety. Once the fetal umbilical vessels change from double UA to SUA and the UA-PI is low, clinicians may consider the possibility of UA thrombosis and embolism. However, more relevant clinical data are required to investigate the safety of low-PI SUA and its clinical management strategies.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the

work.

Concept and design: Ruiman Li, Yanting Lin, Jie Chen, Andong He, Ruiling Yan

Critical review of the manuscript for important intellectual content: Ruiman Li, Jie Chen, Andong He

Supervision: Ruiman Li

Acquisition, analysis, or interpretation of data: Yanting Lin, Jie Chen, Andong He, Ruiling Yan

Drafting of the manuscript: Yanting Lin, Ruiling Yan

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

Acknowledgements

Yanting Lin and Ruiling Yan contributed equally to the work and should be considered co-first authors. Data are available on reasonable request. The data are stored as de-identified participant data which are available on request to Yanting Lin(linyantingjnu@126.com).

References

1. Li TG, Wang G, Xie F, et al.: Prenatal diagnosis of single umbilical artery and postpartum outcome . Eur J Obstet Gynecol Reprod Biol. 2020, 254:6-10. 10.1016/j.ejogrb.2020.08.047
2. Ren J: Sonographic diagnosis and clinical significance of umbilical arterial atresia . Saudi Pharm J. 2017, 25:482-7. 10.1016/j.jsps.2017.04.010
3. Friebe-Hoffmann U, Hiltmann A, Friedl TW, Lato K, Hammer R, Janni W, Kozlowski P: Prenatally diagnosed single umbilical artery (SUA) - retrospective analysis of 1169 fetuses. Ultraschall Med. 2019, 40:221-9. 10.1055/s-0043-123463
4. Li H, Qufeng W, Wei W, Lin X, Zhang X: Umbilical artery thrombosis: two case reports . Medicine (Baltimore). 2019, 98:e18170. 10.1097/MD.0000000000018170
5. Baron J, Weintraub AY, Sciaky Y, Mastrolia SA, Speigel E, Hershkovitz R: Umbilical artery blood flows among pregnancies with single umbilical artery: a prospective case-control study. J Matern Fetal Neonatal Med. 2015, 28:1803-5. 10.3109/14767058.2014.968845
6. Alhousseini A, Jaiman S, Hernandez-Andrade E, Zeineddine S, Qureshi F, Jacques SM: Umbilical artery thrombosis with associated acute and severe fetal growth restriction and transient severe protein S deficiency: report of a case with prenatal ultrasound diagnosis allowing for timely intervention and good outcome. Case Rep Obstet Gynecol. 2018, 2018:6324362. 10.1155/2018/6324362
7. Avagliano L, Marconi AM, Candiani M, Barbera A, Bulfamante G: Thrombosis of the umbilical vessels revisited. An observational study of 317 consecutive autopsies at a single institution. Hum Pathol. 2010, 41:971-9. 10.1016/j.humpath.2009.07.026