

TWO PLACENTAS IN SINGLETON PREGNANCY WITH FUSED UMBILICAL CORD: A CASE REPORT

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Two placentas in singleton pregnancy with fused umbilical cord which has its own placental insertion site forming 3-vessel cord at fetal end is an extremely rare case. This present case describes two placentas with fused umbilical cord with an episode of vanishing twin syndrome and there seems to be a strong relationship between these two events. A 37-year-old woman, gravid 0, para 0, visited emergency room with an episode of vaginal bleeding without pelvic cramps at 8 weeks and 5 days of gestation and repeated ultrasonic exams revealed reabsorption of vanishing twin and two separate placentas on anterior and posterior body of uterus. At 40 weeks and 4 days, the patient delivered a viable female infant weighing 3,900 g via Cesarean section and postpartum examination of the placentas and membranes confirmed two placentas with fused umbilical cord. Two placentas were almost equal in size and there were 2 cord insertions, 1 into each placenta. The cord at each of the placental disc had marginal insertion site and main placental disc cord had 2 arteries with one vein (3 vessel-cord) whereas side placental disc cord had one artery with one vein (2 vessel-cord). Several hypothesis for this two placentas with fused umbilical cord in singleton pregnancy, were proposed including placenta abnormalities after *in vitro* fertilization-embryo transfer procedure, succenturiate lobes and fetus in fetus, however, further evaluation is need.

Keywords: Two placentas; Fused umbilical cord; Vanishing twin syndrome; Succenturiate lobes

Two placentas are rare in pregnancies, including succenturiate placental [1]. Two placentas with fused umbilical cord forming 3 vessels cord at the fetal end which has its own insertion site to each placental disc is an extremely rare case in a singleton pregnancy. In this case, we observed two placentas with fused umbilical cord with an episode of vanishing twin syndrome and there seems to be a strong relationship between these two events. Here, we report two placentas in singleton pregnancy with fused umbilical cord in the pregnant woman of vanishing twin.

Case Report

A 37-year-old woman, gravid 0, para 0, visited emergency room with an episode of vaginal bleeding, diagnosed as threatened abortion with 1.93×1.71 cm sized subchorionic hematoma. The fetus was 8 weeks and 5 days sized which was resulted from *in vitro* fertilization-embryo transfer (IVF-ET) and there was 2.23×1.62 cm (about 6 weeks size) gestational sac like lesion at

uterine fundus (Fig. 1A). Follow-up ultrasonic evaluation revealed previously found gestational sac like lesion which contained no embryo with subchorionic hematoma and this appeared to be vanishing twin syndrome. No additional subchorionic hematoma and empty gestational sac were found at 21 gestational week, however, the placenta appeared equally divided into anterior and

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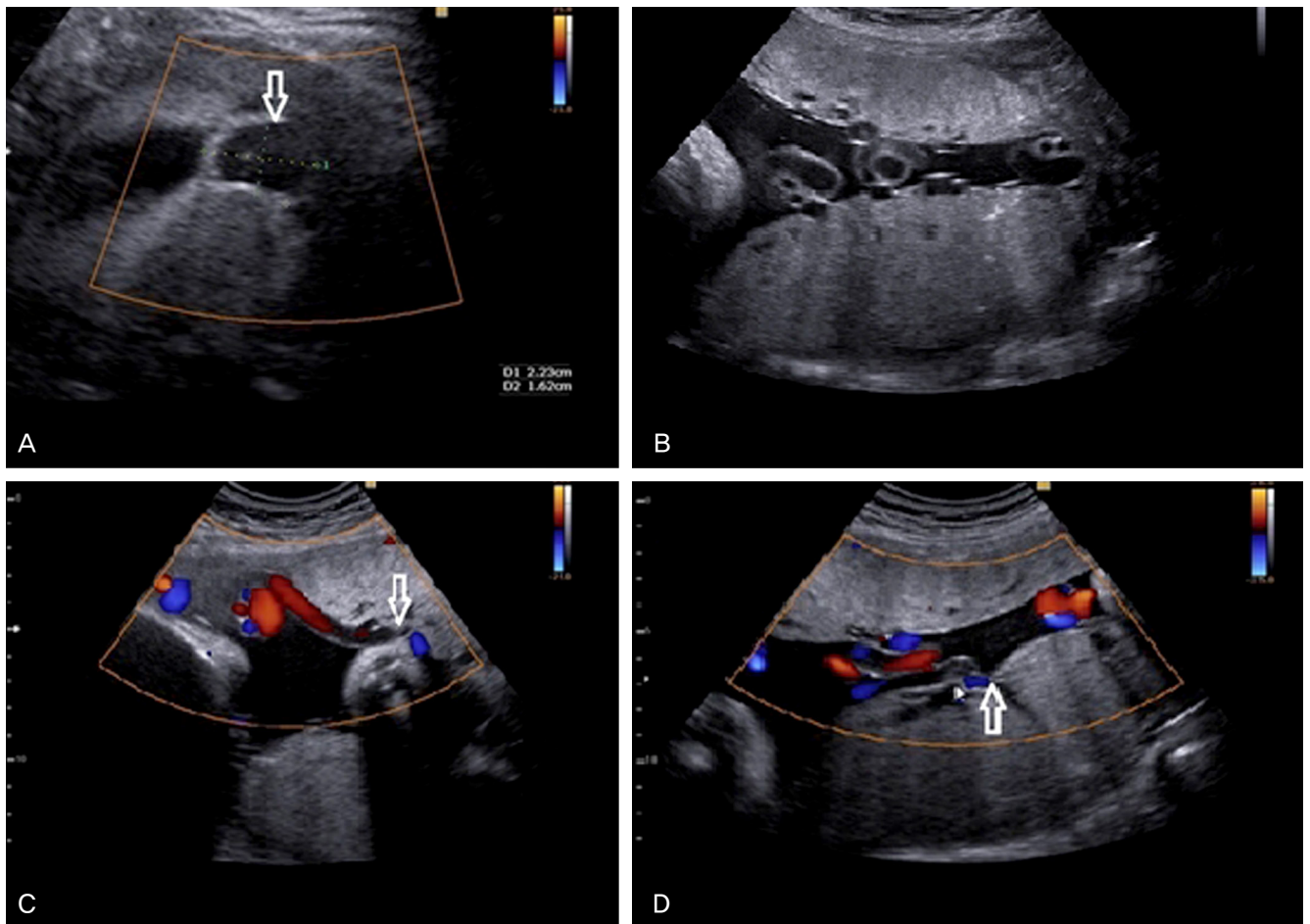


Fig. 1. (A) Vanishing twin syndrome—gestational sac containing no embryo. (B) Two separate placental discs—anterior and posterior, respectively. (C) Side placental disc with cord insertion site (white arrow)—anterior. (D) Main placental disc with cord insertion site (white arrow)—posterior.

posterior lobes with umbilical cord containing 3 vessels (Fig. 1B). Closer inspection with follow-up ultrasonic exam confirmed two placentas—anterior and posterior, respectively—and the cord Doppler systolic/diastolic (S/D) ratio remained in the normal range 1.9 and 2.24, respectively. Among the two placentas, it was difficult to differentiate main placenta which supply fetus because of the individual umbilical cord of each placenta and the complexity of two umbilical cords in amniotic cavity (Fig. 1C, 1D).

At 40 weeks and 4 days, the patient delivered a viable female infant weighing 3,900 g via Cesarean section with diagnosis of suspicious cephalopelvic disproportion and an Apgar score was 9/10 at 1 and 5 minutes. Inspection of the postpartum placentas, membranes, and cord confirmed ultrasound findings. The placenta consists of two placental discs— $13 \times 16 \times 3$ cm sized main disc and $13 \times 12 \times 2.5$ cm sized side disc, respectively—and there were 2 cord insertions, 1 into each placenta (Fig. 2A). The cord at each of the placental disc had marginal insertion site and main placental

disc cord had 2 arteries with one vein (3 vessel cord) whereas side placental disc cord had one artery with one vein (2 vessel cord) (Fig. 2B, 2C). The umbilical cord had total length of 24.5 cm and the umbilical cord of the side placental disc was fused at the insertion site of the main umbilical cord forming normal 3 vessel cord at the fetal end. The fused umbilical cord measured 11 cm in length and 0.8 cm in diameter.

After delivery, the baby was taken whole body X-ray and abdominal ultrasound to exclude fetus in fetus and 2-D echocardiography was also taken to exclude cardiac input overloads resulted from two placentas. No abnormalities were found in the evaluation of the infant (Fig. 2D).

Discussion

Two placentas are rare in pregnancies, including succenturiate pla-

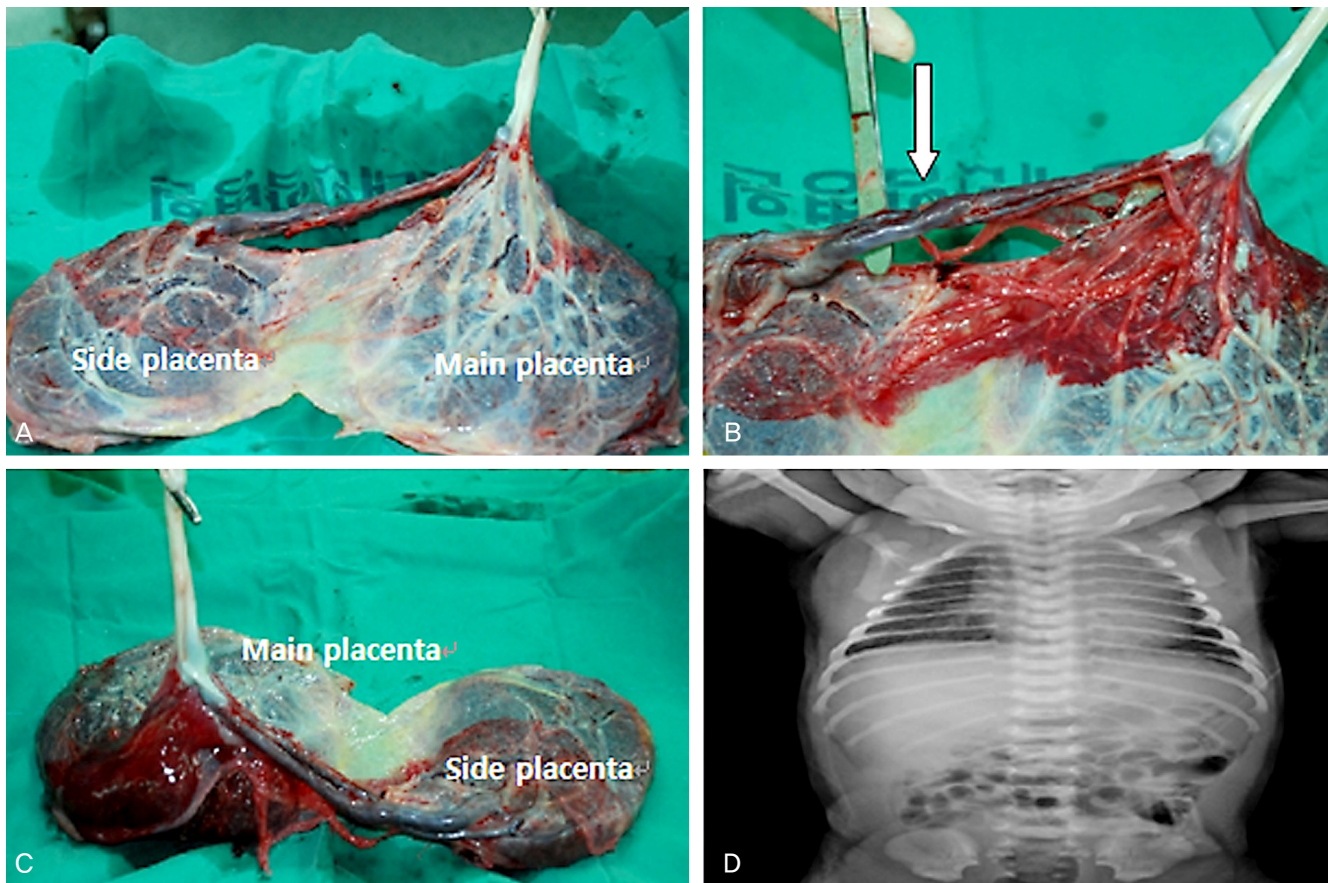


Fig. 2. (A) Two separate placental discs and two individual cord insertions. (B) 2-vessel cord containing one artery and one vein (white arrow) and fusion site. (C) Side umbilical cord fused at insertion site of main placental disc forming 3-vessel cord. (H) Whole body X-ray of the infant—no abnormal findings were found.

cental [1]. Two placentas with fused umbilical cord forming 3 vessels cord at the fetal end which has its own insertion site to each placental disc is an extremely rare case in a singleton pregnancy. Moreover, this present case describes two placentas with fused umbilical cord with an episode of vanishing twin syndrome and there seems to be a strong relationship between these two events. There was only one case of a duplicated placenta and bifurcated umbilical cord in a singleton pregnancy, but this case was not related with vanishing twin [2]. Therefore, this present case may be the first case which describes two placentas with fused umbilical cord that is related with vanishing twin.

Vanishing twin syndrome is the identification of a multiple gestation with subsequent disappearance of one or more fetuses. The rate of multiple gestations at conception is higher than the incidence noted at birth [3]. The frequency of multiple gestations is 3.3% to 5.4% at 8 weeks' gestation [2]. Vanishing twin syndrome occurs in 21% to 30% of multiple gestation [4]. This vanishing

twin case was resulted from IVF-ET with one normal pregnancy and one gestational sac containing no embryo. A relationship between placenta morphologic features and the superficial implantation and/or inadequate orientation of the blastocyst after IVF has been proposed [5]. Considering the fact that this present case was resulted from IVF-ET, these well-known relationships between placenta morphology and IVF-ET could account for the two placentas in this case.

According to classification of placental morphology, two separate placentas of this case could be considered as succenturiate placenta [6] which provide another possible hypothesis for this case. Presenting on ultrasound as a small section of placental tissue distant to the main placental body, they are thought to represent a form of trophotropism of the placenta. It is believed that the placenta recedes from areas of inadequate blood supply such as fibroids and may experience some proliferation of villi on the other placental margins. Thus, trophotropism may result in a separated,

or succenturiate, section of the placenta [2]. Suzuki et al. [7] reported that the incidence of succenturiate lobes of placenta in twin pregnancies was significantly higher than that in singleton pregnancies. Furthermore, in their earlier study with singleton pregnancies, Suzuki and Igarashi [8] also reported the frequency of maternal age >35 years and history of infertility using IVF in patients complicated by succenturiate lobes of placenta were significantly higher than those without succenturiate lobes of placenta. In this present case, vanishing twin syndrome may attribute to this two placentas. However, the diagnosis of succenturiate lobes of placenta requires the additional placental lobe that is much smaller than the largest lobe of placenta macroscopically and the presence of subchorionic vessels between the main placental disk and the accessory lobe confirmed by placental pathologist [2,7]. In this case, the placentas were almost equal in size and placed on both anterior and posterior portion of the uterus. The only connection between the two placental discs was the free-floating, fused umbilical cord without connection of subchorionic vessels. Thus, in this case, a succenturiate lobe could be considered according to the placenta classification, but this special morphology is not match with succenturiate lobes of placenta that could be explained true duplicated placentas.

In vanishing twin syndrome, there may be complete reabsorption of a fetus, formation of a fetus papyraceus, or development of a subtle abnormality on the placenta such as a cyst, subchorionic fibrin, or amorphous material [9]. In this case, repeated antenatal ultrasonic exam revealed complete reabsorption of the gestational sac which contained no embryo. During the process of reabsorption, fetus in fetus, which was first described by Meckel, was thought to be ruled out for continuously developing side placental disc and fused umbilical cord. Fetus in fetus is a rare condition in which a malformed parasitic twin was found inside the body of its partner, usually in the abdominal cavity. It represents an aberration of monozygotic diamniotic twinning [10]. We considered the possibility of fetus in fetus with two placentas. However, in this case, no abnormalities were found during the repeated antenatal ultrasound exams and the findings of whole body X-ray and abdominal ultrasound for the neonate were normal. Furthermore, neonatal 2-D Echocardiography of the infant was also normal.

There are several reports which describes bifurcated umbilical cords in monoamniotic twin gestations [11]. A bifurcated umbilical cord in a monoamniotic twin gestation is an event that takes place when separation of the 2 embryos is delayed until just hours before they would have become conjoined on day 13 postfertilization [1,11]. However, to our knowledge, this present case is the

first case of two placentas with fused umbilical cord which originated from vanishing twin syndrome in singleton pregnancy and further evaluation of etiology and morphologic features is needed.

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References

1. Benirschke K, Kaufmann P. Placental shape aberrations. In: Benirschke K, Kaufmann P, editors. *Pathology of the human placenta*. 4th ed. New York (NY): Springer-Verlag; 2000. p. 300-414.
2. Walkup DW. A rare case of duplicated placenta and bifurcated umbilical cord in a singleton pregnancy. *J Diagn Med Sonogr* 2001;17:280-5.
3. Landy HJ, Weiner S, Corson SL, Batzer FR, Bolognese RJ. The "vanishing twin": ultrasonographic assessment of fetal disappearance in the first trimester. *Am J Obstet Gynecol* 1986;155:14-9.
4. Sampson A, de Crespigny LC. Vanishing twins: the frequency of spontaneous fetal reduction of a twin pregnancy. *Ultrasound Obstet Gynecol* 1992;2:107-9.
5. Jauniaux E, Englert Y, Vanesse M, Hiden M, Wilkin P. Pathologic features of placentas from singleton pregnancies obtained by in vitro fertilization and embryo transfer. *Obstet Gynecol* 1990;76:61-4.
6. Feldstein VA, Harris RD, Machin GA. Ultrasound evaluation of the placenta and umbilical cord. In: Callen P, editor. *Ultrasonography in obstetrics and gynecology*. 5th ed. Philadelphia (PA): Saunders; 2007. p.721-4.
7. Suzuki S, Igarashi M, Inde Y, Miyake H. Abnormally shaped placentae in twin pregnancy. *Arch Gynecol Obstet* 2010;281:65-9.
8. Suzuki S, Igarashi M. Clinical significance of pregnancies with succenturiate lobes of placenta. *Arch Gynecol Obstet* 2008;277:299-301.
9. Landy HJ, Keith LG. The vanishing twin: a review. *Hum Reprod Update* 1998;4:177-83.
10. Chua JH, Chui CH, Sai Prasad TR, Jabcobsen AS, Meenakshi

A, Hwang WS. Fetus-in-fetu in the pelvis: report of a case and literature review. Ann Acad Med Singapore 2005;34:646-9.

11. Fraser RB, Liston RM, Thompson DL, Wright JR Jr. Monoam-

niotic twins delivered liveborn with a forked umbilical cord. Pediatr Pathol Lab Med 1997;17:639-44.

단태임신에서 융합된 단일 탯줄을 가진 두 개의 태반 1예

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박나윤, 류민지, 조금준, 오민정, 김해중, 김 탁, 김선행, 홍순철

단태임신에서 두 개의 독립된 태반과 각각의 태반으로부터 형성된 탯줄이 하나의 탯줄로 융합되어 태아에게 연결되는 것은 매우 드문 현상이다. 37세 초산인 여성이 임신 8주 5일에 vanishing twin syndrome이 의심되는 절박유산으로 입원했으며, 이후 산전 초음파 진찰 결과 자궁벽 전면과 후면에 위치한 두 개의 독립된 태반이 진단되었다. 분만 후 시행한 검사결과 각각의 독립된 태반의 가장자리에서 형성된 탯줄 중 주태반의 탯줄은 2개의 동맥과 1개의 정맥, 부태반의 탯줄은 1개의 동맥과 1개의 정맥으로 이루어져 있었으며 주태반의 탯줄 시작부위에서 하나로 융합되는 형태를 보였다. 단태임신에서 관찰된 두 개의 태반과 탯줄 융합은 여러 가지 기전-체외수정시술, 부태반의 형성, fetus in fetu 등-에 대해 고려할 수 있으나, 향후 좀 더 체계화된 연구가 필요한 실정이다.

중심단어: 두 개의 태반, 탯줄융합, 주태반, 부태반