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## Bilobed placenta and management choices: literature review and two cases studies

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### ABSTRACT

**Objective.** Bilobed placenta is a placental morphological variation with an estimated incidence of 4% of all pregnancies. It is associated with an increased risk of cord insertion anomalies and vasa previa. We searched the literature, focusing on the diagnosis, management, and particular aspects of cases of bilobed placenta. Moreover, we described our experience with two cases of bilobed placenta.

**Materials and Methods.** A literature search was conducted from January 2011 to January 2022. We selected all clinical studies in English, investigating the diagnosis and management of cases of bilobed placenta. Moreover, we reported two cases of a bilobed placenta.

**Results.** Ten papers were included in the review: 7 case reports, 1 cross-sectional study, 2 retrospective analysis. Based on the type of found articles, the quality of evidence is limited, mainly on the pathophysiology and aetiology of this anomaly. Moreover, our search showed a significantly lower incidence compared to the literature. About our reported two cases of a bilobed placenta associated with anomalous cord insertion: case 1, characterized by a velamentous cord insertion, case 2, with a marginal previa placenta associated with vasa previa.

**Conclusions.** A close ultrasound follow-up should be performed in case of suspicion of bilobed placenta to confirm the morphology and proximity to the cervix, assess the umbilical cord's insertion and to identify other eventual associated anomalies. The management should be carried out by a dedicated team of specialists with experience with placenta abnormalities to reduce the risk of adverse foetal and maternal outcomes.

## OBJECTIVE

The endometrial environment plays a key role in the physiological implantation [1, 2] and development of the placenta vascular framework, which can be impaired and lead to placenta-related obstetric diseases [3, 4].

A bilobed placenta (or bipartite placenta) is a placental morphological variation and refers to a placenta separated into two nearly equal lobes. The estimated incidence is about 4% of all pregnancies [5]. The mechanism of formation and the causes remain unknown; the leading hypothesis is that localized atrophy is possible due to poor decidualization and vascularization in a section of the uterus [5-7].

In the case of the bilobed placenta, the umbilical cord may insert itself into one of the lobes: indeed, it is associated with a higher incidence of cord insertion anomalies; insertion in both lobes, velamentous insertion or insertion between the lobes. Moreover, vasa previa, a complication in which foetal blood vessels cross near the internal uterine orifice, represents a rare but possibly severe obstetrical issue with an increased risk of adverse foetal outcomes; rupture of vasa previa often results in foetal haemorrhagic shock and death.

Although there is no increased risk of associated foetal abnormalities, the bilobed placenta may be complicated by first trimester bleeding, polyhydramnios, placental abruption and retention.

It has been documented that a diagnosis of bilobed placenta can be performed in the first trimester, with a close correspondence at the time of delivery [8-10]. Therefore, early diagnosis of anomalous placenta is crucial for patient management, mainly regarding delivery timing.

In the current study, we aimed to analyse international literature on the topic, focusing on the diagnosis, management, and particular aspects of cases of the bilobed placenta. Moreover, we describe our experience with two cases of bilobed placenta, managed differently based on the characteristics of cord insertion.

## MATERIALS AND METHODS

The research was conducted using the following electronic databases: Medline, Embase, Web of science, ClinicalTrials.gov, Cochrane Library and Scopus. The studies were identified using combinations

of the search terms “bilobed placenta”, “bipartite placenta”, “placenta anomalies”, “placenta”, “ultrasoundography” and “pregnancy”, from January 2011 to January 2022; we chose to limit the research to the time span of the previous decade to focus research on recent and updated data. The current study conforms to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) [11].

We selected all clinical studies investigating the diagnosis and management of cases of bilobed placenta (prospective observational studies, retrospective studies, case-control studies, case series and case reports). Studies were excluded as follows: 1) review or meta-analysis that did not report their data; 2) abstracts and extracts from congresses; 3) animal studies; 4) no primary data or incomplete data; 5) duplicate data. Only articles in the English language were included in the search. All review stages were conducted independently by two reviewers (G.C. and S.P.), who assessed the electronic search, study eligibility, inclusion criteria, data extraction, and data analysis. Disagreements were resolved by discussion with a third reviewer (A.S.). All relevant data of the included articles are reported and commented on, with particular attention to the management of the described cases and primary outcomes. Moreover, we reported our experience with two cases of bilobed placenta with anomalous cord insertion. An informed consent was obtained in accordance with Declaration of Helsinki.

## RESULTS

Our search returned 1,024 records; among these, 855 were retained after removing duplicates. We screened the titles and abstracts and excluded 845 ineligible records, and finally 10 papers were included in our review (shown in **Figure 1**). These 10 studies included 5 case reports [8, 9, 12-14], 2 case series [15, 16], 1 cross sectional study [17], and 2 retrospective studies [18, 19]. One case report [12], 1 case series [13] and 1 retrospective study [19] dealt with bilobed placenta in twins.

Regarding the cases of our experience, both were diagnosed and managed at the referral centre of “Villa Sofia Cervello” Hospital in Palermo, in the period between 2020 and 2021.

We did not use a systematic approach in reporting results due to the limited quantity of literature on this particular placental condition; therefore, we conducted a narrative review and reported the

most relevant studies to provide the reader with an exhaustive and updated overview of the main aspects of the topic. For the same reasons, no statistical analysis was performed.

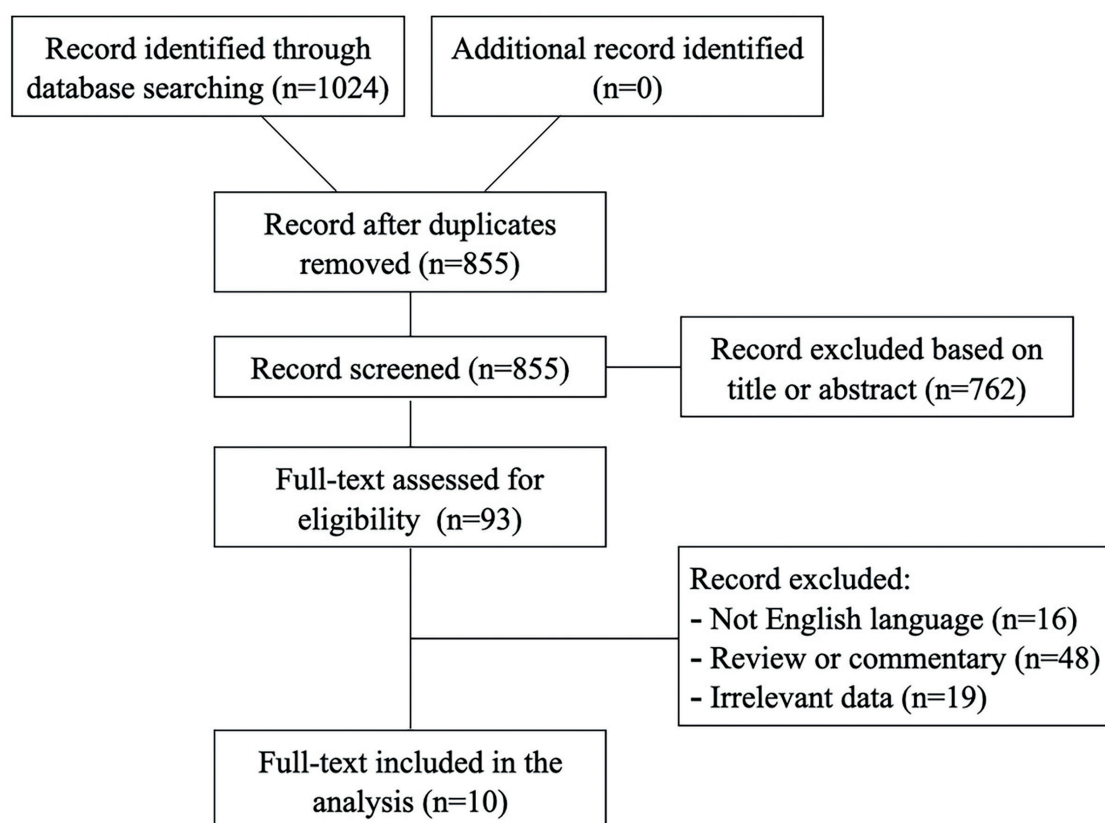
### Case reports

All the relevant data of the case report included in the present review are described in **Table 1**. In 2014, Brighthouse *et al.* reported, for the first time, a case of bilobed placenta associated with a transamniotic vascular connection, which resulted, on successive examination, in an area of thin attenuated placenta measuring  $12 \times 7$  cm [14]. In this case, an elective successful caesarean section was chosen, as the vascular connection was thinning (with restricted foetal movement) and the presentation was breech; however, no congenital malformation or other foetal anomalies were identified [14]. In 2015, Ukwenya *et al.* described a rare case of 'H' bilobate placental partition, which created the impression of two separate gestational sacs and the foetus was located in one of these; the foetus suffered the risk of oligohydramnios and insufficient space due to the partitioning of the uterine cavity. The Authors correlated this con-

dition to the perinatal complication of intrauterine growth restriction (IUGR) [13].

Biswas *et al.* reported an early diagnosis of a case of bilobed placenta previa, successfully managed with serial scanning and successive caesarean delivery; post-delivery placental pathology also identified a velamentous cord insertion [9]. More recently, Dabkowska *et al.* presented a rare case of bilobed placenta with marginal cord insertion (also defined "battledore cord insertion") without associated foetal anomalies [8]; based on their experience, the authors decided on an elective caesarean section due to abnormal placenta formation and the increased risk of foetal complications (*e.g.*, risk of vascular rupture and foetal haemorrhage during labour).

Kutuk *et al.* in 2020 focused on the importance of suspect monozygosity in the case of two separate placentas (bilobed placenta) with the ultrasound T-sign (direct binding of the two thin amniotic membranes) in same-sex twins [12]. Although conventionally the presence of two separate placentas is regarded as a marker of dichorionic twin, the Authors reported an unusual case of bipartite monochorionic twins, as confirmed by microscopic and DNA examinations [12].



**Figure 1.** PRISMA flow diagram of studies identified in the systematic review.

**Table 1.** Main data of the case reports included in the review.

| Reference        | Patient's age (parity) | Type of pregnancy | Symptoms/signs antepartum | Placenta site               | Antepartum diagnosis  | Time of delivery (weeks) | Postpartum diagnosis           | Cord insertion                        |
|------------------|------------------------|-------------------|---------------------------|-----------------------------|---|--------------------------|--------------------------------|---------------------------------------|
| Brighthouse 2014 | 30                     | Singleton         | 4 episodes of haemorrhage | Fundus-posterior wall       | Transamniotic vascular connection from uterine fundus to possible large anterior succenturiate lobe | CS (34)                  | Bilobed placenta               | On smaller lobe                       |
| Ukwenya 2015     | 31                     | Singleton         | IUGR                      | Fundus                      | H-shaped placenta (middle vertical placenta portion between the two lobes)                          | CS (36)                  | "H" bilobed placenta           | Eccentric at 3 o'clock position       |
| Biswas 2016      | 23                     | Singleton         | Moderate IUGR             | Central placenta previa     | Bilobed placenta previa   | CS (36)                  | Bilobed placenta previa        | Velamentous, over the os              |
| Dabkowska 2020   | 32                     | Singleton         | None                      | Anterior and posterior wall | Bilobed placenta  | CS (38)                  | Bilobed placenta               | Marginal                              |
| Kutuk 2020       | 35 (2)                 | Twins             | None                      | Anterior and posterior wall | Monochorionic diamniotic pregnancy  | CS (37)                  | Bilobed monochorionic placenta | Central and paracentral, respectively |

IUGR: intrauterine growth restriction; CS: caesarean section.

### Case series

Castejon *et al.* described two cases of bilobed placenta seen in two pregnancies at 37 and 38 weeks of gestation, with two live births, focusing on the examination of the villous tree with a light microscope and to study the histomorphology of the lobes (two small lobes were found in one placenta and other in the second placenta) [16]. Degenerative changes at the level of vessels of the placental villi were noted in stem villi: stromal lysis, multiple capillarity, vascular congestion, and increased dilatation of vessels. All the evidence (regions of immature villi and preinfarction, deficiency of terminal villi in mature intermediate villi, destroyed villi) indicated extensive hypoxic villous damage [16]. In 2019, Abgral *et al.* reported two cases of a spontaneous monochorionic diamniotic pregnancy with an anomaly consisting of two distinct placental masses diagnosed at 12 weeks of gestation [15]. In the first case, a 31-year-old patient, the pregnancy was uncomplicated, and it ended with spontaneous labour and the birth of two healthy newborns. Macroscopic analysis of the placenta showed two distinct placental masses, each with a marginal cord insertion. The second case, a 36-year-old patient, was characterized by twin-to-twin transfusion syndrome (TTTS) at 19 weeks of gestation, which was resolved by selective foetoscopic laser photocoagulation of the anastomosis; follow-up ultrasound scans showed two eutrophic foetuses, normal amniotic fluids and Dopplers. After induction of labour at 36 weeks of gestation, the patient delivered vaginally. Macroscopic analysis showed a multilobed placenta (bilobed placenta

with associated an aberrant cotyledon) and cord insertions that were marginal for twin A and velamentous for twin B. In both reported cases, histological analysis of the interamniotic membrane found no interposition of the chorion in interamniotic membranes, confirming the diagnosis of a monochorionic diamniotic pregnancy.

### Cross-sectional studies

Reddy *et al.* conducted a cross-sectional study in 975 cases (all pregnancies of > 32 weeks of gestation, between April 2012-June 2013) to evaluate the prevalence and pattern of placental and umbilical cord abnormalities [17]. Data on maternal and foetal outcomes were not reported. Immediately after delivery, all placental discs were examined for insertion of chorioamniotic membranes, umbilical cord insertion and extra lobes. A total of 262 different abnormalities (26.87%) occurred, and 82 (8.41%) were placental; in particular, 11 cases of bilobed placenta were identified (3.76% among total abnormalities) and only in one case did the cord have a velamentous insertion.

### Retrospective studies

The retrospective analysis (study period 2014-2019) of Walter *et al.* aimed to identify possible implications and risk factors in monochorionic twin pregnancies having a bilobed placenta; monochorionicity was confirmed either by the presence of a T-sign (one chorion and two separate amnion cavities) or by histological examination postnatally [19]. Bipartite placenta



was diagnosed when two placenta lobes were identified with no placental connection. Among five twin pregnancies included in the study, three were complicated by TTTS, one by selective IUGR and one by severe foetal malformations affecting the central nervous system; foetoscopic laser coagulation of vascular anastomoses was performed for TTTS and in one case, a selective termination of pregnancy was carried out (selective IUGR). The overall survival rate at birth was 90% with a neonatal survival rate of 66.63% (6 out of 9 newborns). Monochorionicity was confirmed in all the cases. Umbilical cord insertion was abnormal in 6 cases: 2 marginal and 4 velamentous.

A recent retrospective cohort study by Volodarsky-Perel *et al.* (study period 2009-2017) was conducted to evaluate the effect of foetal gender on the placental histopathology pattern and perinatal outcomes in singleton live births resulting from IVF treatment; primary outcomes included evaluation of placental features, and secondary outcomes included foetal, maternal, perinatal and delivery complications [18]. After adjustment for potential confounding factors, the female gender was significantly associated with bilobed placenta (OR 0.2; 95%CI 0.06-0.8); the prevalence of adverse foetal and maternal outcomes was similar between the groups.

## Our experience

### First case

The first case is a 27-year-old patient, primigravida, with no comorbidities or history of surgical procedures. She was diagnosed with a possible placenta anomaly in a singleton pregnancy during the first trimester ultrasonographic evaluation, performed in a private clinic. So, the patient was referred to our centre at 13 weeks gestational age,

and a placental ultrasound revealed a bilobed placenta corresponding with the right lateral wall, with the cord insertion between the two lobes. This insertion type is defined as velamentous (**Figure 2a**). The cord had two umbilical arteries, one umbilical vein and a normal coil index; the foetal parameters corresponded to gestational time.

Foetal growth and doppler were normal over the following weeks. Based on these findings, we decided on wait management with close obstetric follow up. A dedicated multidisciplinary team decided for spontaneous birth, also considering the placental position, aspects, and development. Induction of labour was not considered.

The patient delivered spontaneously at 38 weeks and 6 days. The baby was born with no complications and weighed 3,100 g at birth, with no apparent congenital malformation.

The placenta was removed intact after the delivery, although manual removal was prolonged. On examination, the placenta appeared bilobed (one lobe slightly larger than the other) with perfectly separated lobes; the disc outline showed no evidence of placental membranacea, percreta, increta or accreta. The diagnosis of velamentous cord insertion was confirmed (**Figure 2b,c**): the membranes contained a 12 cm placental free segment with suspended velamentous vessels that connected the two lobes. Mother and baby were discharged from the hospital after two days.

### Second case

The second case is a 32-year-old patient, secundigravida and no history of diabetes mellitus, hypertension, or anaemia. Previous pregnancy was normal with spontaneous delivery at 39 weeks and 5 days. At 12 weeks, the ultrasound evaluation at our



**Figure 2.** Sonographic (a) and post-partum images (b,c) of bilobed placenta of Case 1.

In (a) colour Doppler highlighted the cord insertion between the two placental lobes on the uterine fundus; (b) and (c) images confirmed the placenta anomaly and the velamentous insertion of the cord. Pl: placenta.

referral centre showed a bilobed placenta localized in the lower part of the uterus. The second trimester scan confirmed the diagnosis of a bilobed previa marginal placenta; moreover, the cord insertion was placed between the two lobes, close to the internal uterine orifice with a suspicion of vasa previa. Given the above, a series of ultrasounds was scheduled. Vasa previa were detected as tubular or round structures overlying the cervix and positive to color Doppler (**Figure 3a,b**); pulsed Doppler showed a foetal heart rate confirming the origin of these vessels.

Given the above suspicion and the risks associated with vaginal birth, we planned a caesarean section. At 20 weeks, foetal anatomical evaluation was regular with normal foetal growth. The anterior and posterior placental structures appeared to be equal in size. In addition, amniotic fluid and growth were normal in the following scans.

At 36 weeks and 2 days, there was unexpected vaginal bleeding, and an emergency caesarean section was carried out. A healthy baby weighing 2,600 g was successfully delivered. The blood loss was approximately 800 mL, and the patient did not require a transfusion. The placental inspection confirmed the presence of two lobes, almost equal in size, separated from each other, without signs of placental membranacea or invasion, and the presence of vasa previa. The patient's post-operative course was uncomplicated, and she was discharged three days after delivery, in a healthy condition. The infant was discharged eight days after delivery in a healthy condition.

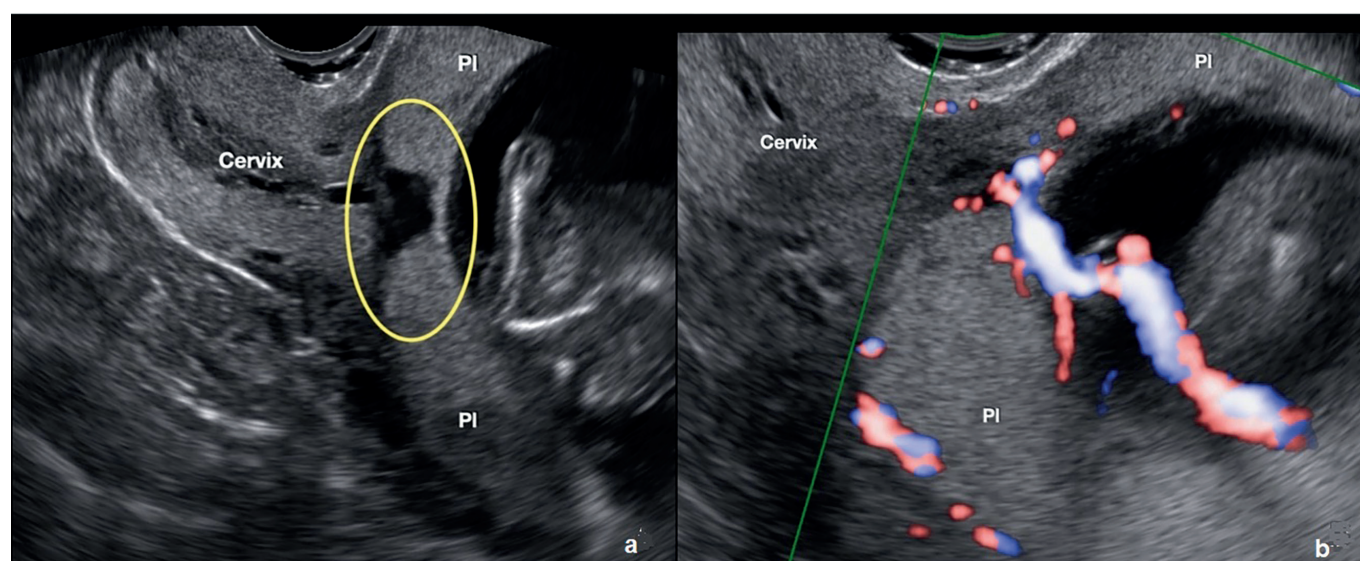
## DISCUSSION

Placental pathologies, placental shape abnormalities, and related maternal and foetal risk factors have been widely discussed in recent years [20-24]. In this paper, we focused on bilobed placenta, to deepen the most controversial aspects of this condition.

Our literature review revealed that, to date, there is not much data available on the pathophysiology and aetiology of this anomaly. Moreover, our search showed a significantly lower incidence compared to the literature. This finding should be taken into account by clinicians regarding the rarity of this placental morphological variation.

Hypothesized risk factors include advanced maternal age, maternal history of infertility, smoking, and diabetes [5, 9, 25-28].

To explain the reasons of the development of a bilobed placenta, three main theories have been formulated. The first theory is based on the failure of the decidua capsularis to regress, persisting as a bipartite placenta [29]. The second suggests the role of insufficient nutrition in the placenta and migration of some parts of it to distant, more favourable areas [30]. The third hypothesis explains the partition of the placenta based on the early implant of the morula on the two adjacent walls of the uterus and then, as the uterus grows and changes its three-dimensional geometry, it pulls apart the separate parts of the placenta [31]. From the clinical point of view, a classification of placental shape deviation origin has been proposed: primarily "active" origin (in the case of an active placental



**Figure 3.** Sonographic images of Case 2.

The yellow ring showed the site of vasa previa between the two placental lobes (a); colour Doppler made the vasa previa clearly evident (b). Pl: placenta.



response to the local environment) and primarily “passive” origin (change of placenta shape due to uterine remodelling) [32].

Although bilobed placenta is associated with antepartum bleeding, polyhydramnios, abruption and retained placenta, literature review data highlighted that it is usually not associated with an increased risk of foetal congenital anomalies [8, 9, 16, 25, 33]; the IUGR associated with the rare form of H-shaped bilobed placenta could be explained by a reduced blood flow related to an impairment of foetal and maternal vascular connections [13].

In the case of bilobed placenta, localization of both the placental lobes and cord insertion appear to be two critical factors in pregnancy management and delivery decision. Although these are rare events, a possible association between central placenta previa and vasa previa should be considered [34, 35]; accurate prenatal diagnosis of vasa previa is crucial: when the condition is not diagnosed antenatally, the perinatal mortality rate is reported to be approximately 44%, whereas 97% of fetuses survive when the diagnosis is performed antenatally [36, 37]. Transvaginal ultrasound and Doppler represent the first diagnostic choice; Magnetic Resonance Imaging could be useful in dubious cases. Further research should investigate strategies for incorporating prenatal screening for vasa previa into routine clinical practice [38]. Given the above, a caesarean section is the most suitable option for diagnosing central placenta previa or vasa previa. Regarding abnormal placental cord insertion, an antenatal serial ultrasound in patients at risk is considered to have a high diagnostic specificity (up to 99-100%); the use of colour Doppler is also useful for this purpose. Velamentous cord insertion is more frequently associated with abnormal shaped placenta, and, in these cases, vaginal delivery could be carefully planned if the patient desires. In our reported experience with a velamentous cord insertion on a bilobed placenta, a spontaneous delivery was considered after close follow up and performed without complications. Of particular interest is the presence of a bilobed placenta in multiple pregnancies. The importance of early determination of chorionicity and amnionity in twin gestations for antenatal and intrapartum management is well known; monochorionic twin pregnancies have an increased risk of adverse perinatal outcomes compared to dichorionic pregnancies [39, 40].

Based on our review, we speculated that the condition of a bilobed placenta in monochorionic twin pregnan-

cies is a rare finding; however, the incidence could be increased compared to the past, mainly due to misdiagnosis of dichorionic pregnancy for the presence of two placental masses [19]. Furthermore, the optimal time for determination of chorionicity in twins is during the first trimester; in the second and third trimester, diagnosis is less accurate and generally not definitive [41]. Thus, in the case of late pregnancy diagnosis, two separate placentas with a T-sign in same-sex twins should raise the suspicion of monozygosity and a close follow-up should be carried out.

## CONCLUSIONS

Prenatal care is paramount for early diagnosis and management of several obstetric conditions [42-44]. An antenatal diagnosis of bilobed placenta may minimize pre- and post-partum complications. Moreover, when a diagnosis of bilobed placenta is suspected, it is crucial to assess the position of the placenta, the umbilical cord insertion and the identification of other associated anomalies such as vasa previa. The key to success in managing these situations is the presence of a dedicated team of specialists such as skilled sonographers, adequate obstetric care, experienced anaesthetist, and neonatologist to ensure a correct follow up and minimize adverse maternal and foetal outcomes.

## COMPLIANCE WITH ETHICAL STANDARDS

### Authors contributions

Gl.Ca.: Conceptualization, writing - original draft. S.P.: Design, writing - review & editing. A.S., F.C.: data acquisition, literature search. Ga.Cu.: literature search, writing - review & editing. V.C.: revising article critically for intellectual content. Gi.Ca., F.D'A: definition of intellectual content, writing - review & editing. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

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### Study registration

N/A.

### Disclosure of interests

The authors declare that they have no conflict of interests.

### Ethical approval

The Institutional Review Board approved the study.

### Informed consent

Informed consent was obtained and signed by patients in accordance with Declaration of Helsinki.

### Data sharing

Data are available under reasonable request to the corresponding author.

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