

A fetiform placental lesion in a monochorionic diamniotic placenta: a diagnostic challenge between teratoma and an acardiac co-twin—a case report^a

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ABSTRACT

Placental teratomas are extremely rare non-trophoblastic tumors of the placenta. A fetiform teratoma is a rare subtype of mature teratoma consisting of highly differentiated tissues that may resemble a malformed fetus but lack true organogenesis, axial skeletal organization and developed systemic anatomical structures. However, it can be difficult to distinguish fetiform-like lesions, particularly those with placental attachment, from a severely regressed or acardiac twin. This case report presents the clinical, macroscopic and histopathological findings of an unusual placental mass, resembling a fetus and containing hair-like structures, which was incidentally detected during a cesarean section in a 26-year-old patient who had had an uneventful antenatal course. Histopathological examination revealed a monochorionic diamniotic placenta containing two umbilical cords, one of which was markedly short, and a lesion measuring approximately 5×5×2.5 cm directly connected to the placenta by the short umbilical cord. Microscopic examination revealed mature tissues derived from multiple germ layers, including skin and skin appendages, adipose tissue, cartilage, striated muscle, ganglionic elements, central nervous system tissue and the choroid plexus. No vertebral axis, long appendicular bones, organized visceral structures or developed systemic anatomical structures were detected. However, the monochorionic diamniotic placental structure and the markedly short second umbilical cord raised the possibility of a severely regressed twin. While the morphological features of the lesion are consistent with a mature fetiform teratoma, a severely regressed or acardiac monozygotic twin could not be definitively ruled out on the basis of morphological findings alone. Without molecular comparative analysis, it is difficult to make a definitive diagnostic distinction between these two possibilities. This case highlights the importance of interpreting morphological findings in conjunction with placental anatomy and, where possible, molecular data when assessing fetiform placental lesions.

Keywords: Placenta, teratoma, fetus in fetu, pregnancy; twin, fetiform placental lesion, monochorionic diamniotic placenta

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INTRODUCTION

Placental tumors are generally classified as trophoblastic and non-trophoblastic lesions.¹ Among non-trophoblastic placental lesions, placental teratomas are extremely rare germ cell tumors; very few cases have been reported in the English-language literature since they were first described by Morville in 1925.^{2–5} They are typically located on the fetal surface of the placenta, between the amnion and the chorion, and are usually detected incidentally after delivery.⁴

A fetiform-type teratoma is an extremely rare subtype of mature teratoma characterized by highly differentiated tissues that may resemble a malformed fetus but lack true organogenesis, a vertebral axis and organized anatomical

structures.⁶ This lesion has historically also been referred to as a ‘homunculus’; however, this terminology remains controversial in modern pathological literature⁷ and, for this reason, the term ‘fetiform-like teratoma’—which is currently accepted—is used throughout this report. Because of its distinctive characteristics, it must be distinguished from fetus in fetu and fetus acardius amorphus, which differ in terms of embryological origin, pathological features and clinical outcomes.^{8,9}

A placental fetiform teratoma is an extremely rare entity, and only a limited number of cases have been described in the literature. A recent systematic review reported 17



histopathologically confirmed fetiform teratomas; however, none of the confirmed cases involved a lesion located in the placenta.⁸ However, in some cases, it is debatable whether placental fetiform-like masses represent a germ cell neoplasm or a severely regressed or acardiac twin; in making this distinction, molecular genetic comparison and genotyping of the lesion against the placenta or the healthy twin, in addition to morphological assessment, may contribute to the diagnostic evaluation.¹⁰ It is therefore important to evaluate morphological findings in conjunction with placental anatomy, clinical features and, where possible, molecular comparative data.

This study presents an unusual fetiform-like placental lesion detected in a monochorionic diamniotic placenta, the morphological features of which are consistent with a fetiform-like teratoma, although the possibility of a severely regressed acardiac twin must also be considered. In addition to the clinical and histopathological characteristics of the case, the diagnostic distinction between a fetiform teratoma and a regressed/acardiac twin, as well as the limitations of this distinction, are discussed in the light of the current literature.

CASE REPORT

A 26-year-old patient, gravida 5, para 2, with a history of two previous abortions, was admitted to our clinic at 38 weeks and 5 days' gestation for an elective cesarean section following two previous cesarean deliveries. No abnormalities were detected during antenatal care, and the pregnancy had been monitored without complications at an external center. The patient first presented at our clinic at 12 weeks' gestation; at that time, the ultrasound scan revealed no findings suggestive of a twin pregnancy. The first-trimester combined screening was normal; the PAPP-A level was 2.63 mIU/ml (1.53 MoM), the free beta-hCG level was 64.4 ng/ml (1.7 MoM), nuchal translucency was measured at 0.9 mm and the crown-rump length at 57 mm, and the combined risk was calculated to be less than 1 in 10,000. The second-trimester detailed anomaly ultrasound scan was also normal. At her first appointment, the patient was 165 cm tall and weighed 80 kg (BMI 30 kg/m²); her total weight gain during pregnancy was 10 kg; she did not smoke and the pregnancy had occurred spontaneously. The placenta was anteriorly situated.

Although the pregnancy had been monitored at an external center as an apparently uncomplicated pregnancy, a pathological examination of the delivered placenta revealed a monochorionic diamniotic placental structure and two umbilical cords, one of which was markedly short.

During a cesarean section performed under spinal anesthesia, a live baby girl weighing 3150 g and measuring 50 cm in length was delivered, with Apgar scores of 9 and 10 at 1 and 5 minutes respectively. Following the removal of the placenta and membranes, macroscopic examination revealed an irregular-appearing mass containing hair-like structures adjacent to the placenta (Figure 1). The material was subsequently sent to the pathology laboratory for histopathological examination.

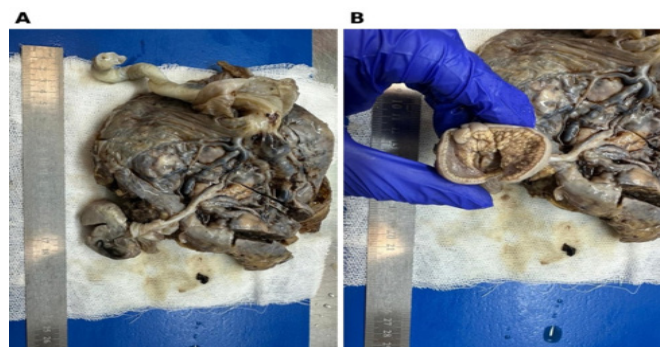


Figure 1. Gross and cut-surface appearance of the fetiform placental lesion. A. Gross appearance of the lesion, showing an irregular nodular mass adjacent to the fetal surface of the placenta with a fetal skin-like external appearance. B. Cut surface of the lesion, revealing a soft, heterogeneous, yellow-tan appearance without gross evidence of organogenesis or organized anatomical structures

Macroscopic pathological examination revealed that the placenta measured 19×15×6 cm and contained two umbilical cords, measuring 27 cm and 6.5 cm in length respectively. The monochorionic diamniotic configuration was confirmed by histological examination of the dividing membrane between the cords, which showed two apposed amniotic layers without intervening chorionic tissue (Figure 2B). In sections taken from the short umbilical cord, a single umbilical artery and a single umbilical vein were observed (Figure 2A). Continuity of the cord vessels into the lesion and placental vascular anastomoses were not specifically documented in the available pathology report. A nodular mass measuring approximately 5×5×2.5 cm and bearing hair follicles on its surface was directly connected to the placenta by the hypoplastic umbilical cord, which measured 6.5 cm in length and 0.5 cm in diameter. The lesion was soft in consistency, cream-pink in color and had a surface appearance resembling fetiform skin. No distinct organ development or organized anatomical structure was observed on macroscopic examination.

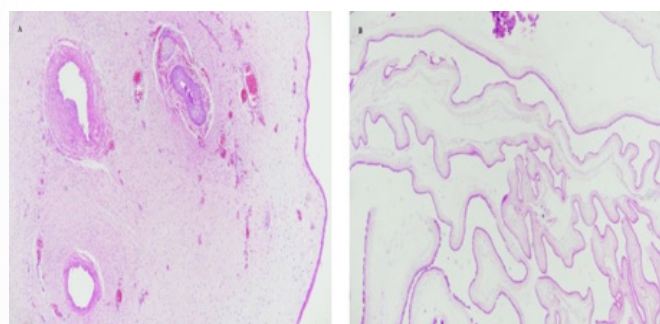


Figure 2. Histopathological findings of the umbilical cord and placental membranes. A. Cross-section of the short umbilical cord demonstrating a single umbilical artery and one umbilical vein (H&E×40). B. Monochorionic diamniotic dividing membrane showing two apposed amniotic layers without intervening chorionic tissue (H&E×40)

Microscopic examination revealed skin and skin appendages, subcutaneous adipose tissue and cartilage within the lesion (Figure 3). In addition, areas of skeletal muscle, ganglionic elements and central nervous system tissue, as well as choroid plexus-like structures, were identified (Figure 4). No vertebral axis, long appendicular bones, organized visceral structures or developed systemic anatomical organization were observed.

Areas of infarction, foci of dystrophic calcification, perivillous fibrin deposits and increased syncytial nodes were also

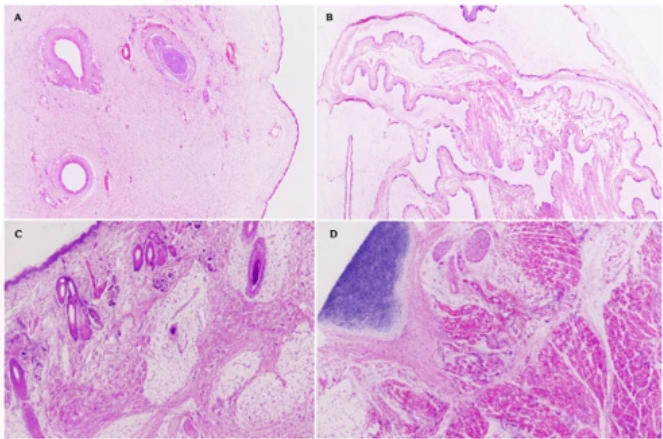


Figure 3. Histopathological features of the fetiform placental lesion. A. Mature skin with adnexal structures and underlying adipose tissue. B. Mature adipose tissue, brown adipose tissue, skeletal muscle, and scattered ganglionic elements within the lesion. C. Coexisting mature white adipose tissue and brown adipose tissue. D. Mature cartilage and skeletal muscle bundles (H&E×100 for all panels). H&E: Hematoxylin and eosin.

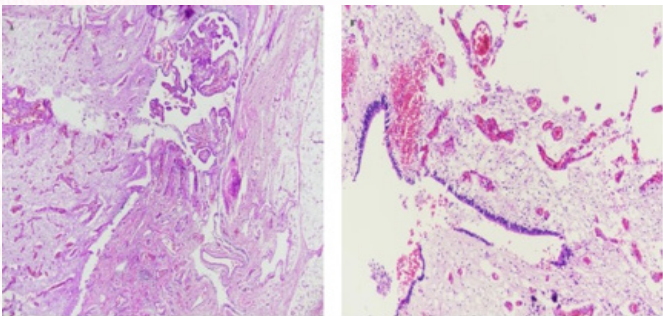


Figure 4. Neural components within the fetiform placental lesion. A. Mature neural tissue with choroid plexus-like structures and adjacent fibroadipose tissue. B. Central nervous system-type tissue showing cerebellar differentiation (H&E×100 for both panels). H&E: Hematoxylin and eosin.

observed in the placental tissue. The morphological findings were assessed as consistent with a fetiform-like placental lesion. However, a definitive distinction between a mature fetiform-like teratoma and a severely regressed or acardiac twin could not be made on the basis of clinical and histopathological findings alone. The timeline is shown in [Table](#).

DISCUSSION

Placental teratomas are extremely rare non-trophoblastic placental lesions; since Morville's initial description in 1925,

only a limited number of cases have been reported in the literature.^{1,2,4} The fetiform variant is even rarer. In a recent systematic review, only 17 fetiform-type teratomas meeting strict morphological criteria were identified.⁸ In this context, the present case represents an unusual fetiform-like placental lesion developing in a monochorionic diamniotic placenta and is of particular interest in terms of the diagnostic differentiation between a placental teratoma and a severely regressed or acardiac twin.

Fetiform teratomas are highly differentiated, mature teratomas that, macroscopically, may resemble a malformed fetus but lack true organogenesis and the regular anatomical organization characteristic of a fetus.^{3,6,9} Histopathological examination of our case revealed mature tissues derived from different germ layers, including skin and skin appendages, adipose tissue, cartilage, skeletal muscle, ganglionic elements, central nervous system tissue and the choroid plexus. No vertebral axis, long appendicular bones, organized visceral structures or immature neuroepithelial elements were observed. These features are consistent with a mature fetiform teratoma in terms of traditional morphological criteria. However, particularly in fetiform lesions with placental involvement, these morphological features alone may not definitively indicate a neoplastic origin.

The etiology of placental teratoma remains unclear. Proposed mechanisms include the aberrant migration of primordial germ cells, abnormal differentiation of germ cells, and developmental processes associated with abnormal twinning.^{1,4,9} The aberrant migration of primordial germ cells is one of the main hypotheses explaining why placental teratomas are classified as germ cell neoplasms. However, the presence of a markedly short second umbilical cord in the present case suggests that the possibility of a severely regressed twin or an acardiac twin in a monochorionic diamniotic placental environment should also be considered.

The main diagnostic challenge in this case is determining whether the lesion represents a true germ cell neoplasm or a monozygotic twin that has regressed to a significant extent. The absence of a vertebral axis, long appendicular bones and organized visceral structures is interpreted in favor of a fetiform teratoma according to traditional morphological

Table. Timeline of the clinical course and diagnostic evaluation		
Timepoint	Clinical event	Findings
12 weeks of gestation	First evaluation at our institution	Ultrasonography showed a singleton pregnancy with no evidence of a second gestational sac or embryo. First-trimester combined screening was low risk.
Second trimester	Detailed fetal anomaly examination	No fetal structural abnormalities were detected.
38 weeks+5 days	Admission for elective cesarean delivery because of previous cesarean sections	Pregnancy had remained clinically uncomplicated.
At delivery	Cesarean section under spinal anesthesia	A live female infant weighing 3150 g was delivered, with Apgar scores of 9 and 10 at 1 and 5 minutes, respectively.
Immediately after delivery	Gross examination of the placenta	An irregular, hair-bearing nodular mass adjacent to the fetal surface of the placenta was identified. The placenta contained two umbilical cords, one of which was markedly short.
Histopathological examination	Evaluation of the placenta, membranes, umbilical cords, and fetiform lesion	Monochorionic diamniotic placental membranes were confirmed. The short cord contained a single umbilical artery and one umbilical vein. The lesion contained mature tissues from multiple germ layers, without a vertebral axis, appendicular long bones, organized viscera, or developed systemic anatomical structures.
Postoperative period	Maternal and neonatal follow-up	The postoperative course of both the mother and the newborn was uneventful, with no maternal or neonatal complications observed.

criteria, while the presence of a monochorionic diamniotic placental structure and a separate, markedly short second umbilical cord supports the possibility of a twin-origin developmental process. The fact that a second gestational sac or embryo was not visualized on the ultrasound scan at 12 weeks' gestation does not definitively rule out the possibility of a monozygotic twin that was lost at an earlier stage or has regressed significantly. Therefore, in the present case, it is difficult to make a definitive distinction between these two entities based solely on the absence of fetiform organization or axial skeletal elements.

Limitations

The limitations of this distinction have also been demonstrated at the molecular level by McHenry et al.¹⁰ The authors carried out DNA genotyping on a lesion that could be assessed as consistent with a placental teratoma according to traditional morphological criteria, and demonstrated that the lesion and normal placental tissue shared largely the same genetic profile at the microsatellite loci examined. In light of these findings, the lesion was re-evaluated as a monozygotic acardiac/amorphous twin, and the importance of ruling out a monozygotic twin origin at the molecular level in the diagnosis of placental teratoma was emphasized. The fact that no molecular comparative analysis was performed in the present case limits the ability to definitively distinguish between a teratomatous origin and the possibility of a severely regressed monozygotic twin.

Short tandem repeat (STR) analysis, or other informative molecular genetic methods, carried out on the lesion, normal placental tissue and, where possible, the healthy twin, may provide further evidence regarding the origin of the lesion. Such investigations can also be carried out on tissue that has been properly fixed in formalin and embedded in paraffin. However, molecular results must also be interpreted with caution; the genetic composition of germ cell teratomas is not homogeneous, and heterozygous genetic features may be preserved in some mature teratomas. It is therefore more appropriate to evaluate molecular findings in conjunction with morphological, clinical and placental findings.

The fundamental significance of this finding lies not so much in its clinical outcomes as in the fact that it highlights the diagnostic difficulties encountered in the classification of placental fetiform-like lesions. Particularly in cases where a second umbilical cord is present in a monochorionic placenta, the diagnosis of a fetiform teratoma should not be based solely on morphological features; the possibility of severe regression or an acardiac monozygotic twin should also be considered. The evaluation of morphological, clinical and placental findings, together with molecular genetic data where possible, may contribute to a more accurate classification of the etiology of these rare lesions.

CONCLUSION

This case presents an unusual fetiform-like placental lesion detected in a monochorionic diamniotic placenta. The absence of a vertebral axis, long appendicular bones and organized visceral structures, coupled with the presence of mature but disorganized tissues originating from different germ layers, is

consistent with a mature fetiform-like teratoma according to traditional morphological criteria. However, the presence of a markedly short second umbilical cord and a monochorionic diamniotic placental structure necessitates consideration of the possibility of severely regressed or acardiac monozygotic twins. The lack of molecular comparative data limits the ability to definitively distinguish between these two possibilities. It is therefore important, when assessing fetiform-like placental lesions, to interpret morphological findings in conjunction with placental anatomy, clinical features and, where possible, molecular genetic data.

ETHICAL DECLARATIONS

Informed Consent

Written informed consent was obtained from the patient for publication of the clinical details and accompanying images.

Peer Review Process

This report underwent external peer review.

Conflict of Interest

The authors declare no conflicts of interest.

Financial Disclosure

This case report did not receive any financial support.

Author Contributions

Concept: AGY, SH; Design: AGY; Control: İK; Resources: SH, HÜ; Materials: HÜ; Data Collection and/or Processing: SH; Analysis and/or Interpretation: AGY; Literature Review: FA; Writing The Article: AGY; Critical Review: İK.

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