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Insights into Congenital Body Stalk Anomaly Coupled with Placenta Accreta Conditions: A Case Report

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Patient: Female, 29-year-old
Final Diagnosis: Body stalk anomaly • P2A0, delivery by caesarean section • placenta accreta • post subtotal hysterectomy due to uterine atony
Symptoms: Asymptomatic
Clinical Procedure: Compression suture • subtotal hysterectomy • transperitoneal caaesarean section • wedge resection
Specialty: Obstetrics and Gynecology

Objective: Rare disease

Background: Body stalk anomaly is a rare abdominal wall defect thought to be a consequence of abnormalities in the development of cephalic, caudal, and lateral embryonic folding and defect in closure of the body wall during embryogenesis. Placenta accreta spectrum (PAS) is a general term frequently used to encompass accreta, increta, and percreta conditions. This report describes a distinct pregnancy with a body stalk abnormality and PAS.

Case Report: A 34-year-old woman, gravida 2 para 1, with no previous abortions, was referred to the Maternal-Fetal Medicine Unit for further investigation of omphalocele at 29 weeks of gestation. Although the defect was not suspected during the first trimester scan, subsequent obstetric ultrasounds revealed a severe abdominal wall defect, kyphoscoliosis, a very rudimentary umbilical cord, and limb defects. Ultrasound examination of the placenta showed increased vascularity at the placental bed and loss of the retroplacental-myometrial radiolucent interface, leading to diagnosis of suspected body stalk anomaly, with PAS. Cesarean delivery was performed at 30 weeks, with plan for conservative treatment for PAS, including uterine-sparing surgery. Baby was born weighing 800 g and measuring 25 cm in length, with an APGAR score of 1-1. Clinical examination confirmed a very short umbilical cord and severe abdominal wall and limb defects. However, due to significant hemorrhage during surgical procedure, cesarean hysterectomy was done.

Conclusions: The management of body stalk anomaly with PAS is challenging. Preconception counseling is important to detect abnormalities earlier, and a multidisciplinary care team is needed to create patients' treatment plans. This congenital defect is invariably fatal.

Keywords: Cesarean Section • Congenital Abnormalities • Placenta Accreta

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Introduction

Body stalk malformation is a very severe and rare congenital anomaly [1]. The body stalk abnormality is the rarest and most severe abdominal wall deformity and is invariably fatal. This is a critical anomaly characterized by the underdevelopment of the abdominal wall, resulting in an open peritoneal cavity that connects to the extra-embryonic coelom, with the fetus remaining linked to the placenta [2]. This uncommon malformation condition has a documented prevalence of 0.12 occurrences per 10 000 births, encompassing live and stillbirths [3]. Potential etiologies of body stalk anomaly include premature amnion rupture leading to direct mechanical pressure and amniotic bands, vascular disruption of the early embryo, and an aberration in the germinal disc [4].

Antenatal screening, particularly standard mid-trimester ultrasound, has greatly improved the detection of significant congenital abnormalities by providing detailed imaging that aids in early and accurate diagnosis [5]. Antenatal diagnosis enables focused diagnostic testing, delivery planning, and the counseling and education of mothers or couples. The instance of body stalk anomaly previously demonstrated no chromosomal abnormalities upon genetic analysis [6,7].

Detailed scan and antenatal diagnosis also allow detection of abnormal placentation, such as placenta accreta spectrum, which is an increasingly common disorder [6,7]. Its severity can vary, and its incidence is rising due to the increasing number of cesarean sections [8]. The co-presence of a severe congenital anomaly with placenta accreta is a very rare combination [6]. Precise prenatal identification facilitates the implementation of optimal treatment, with ultrasound and magnetic resonance imaging (MRI) being the preferred examinations. The primary indications for MRI include ambiguous ultrasound results, the existence of risk factors for placenta accreta/increta/percreta, and a posterior placental position [9].

This case report describes the occurrence of a combination of body stalk anomaly and placenta accreta spectrum. Given the

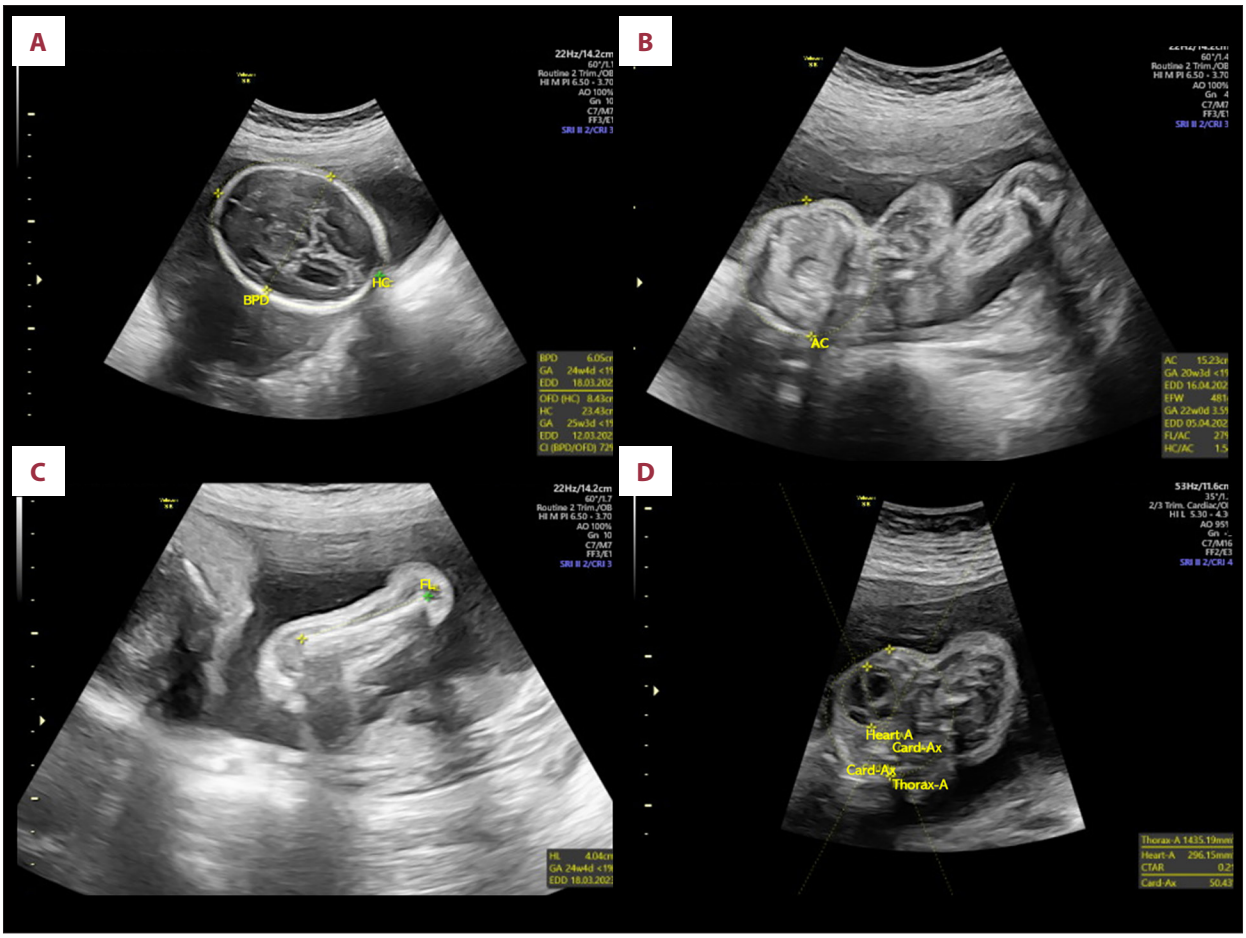

Figure 1. Fetal biometry. (**A**) Fetal head biometry biparietal diameter (BPD) and head circumference (HC) equal to 24 to 25 weeks' gestational age (**B**) abdominal circumference (AC) equal to 20 weeks' gestational age (**C**) femur length (FL) equal to 24 weeks gestational age (**D**) 4-chamber view (4CV) shows cardiac inside thoracic cavity.

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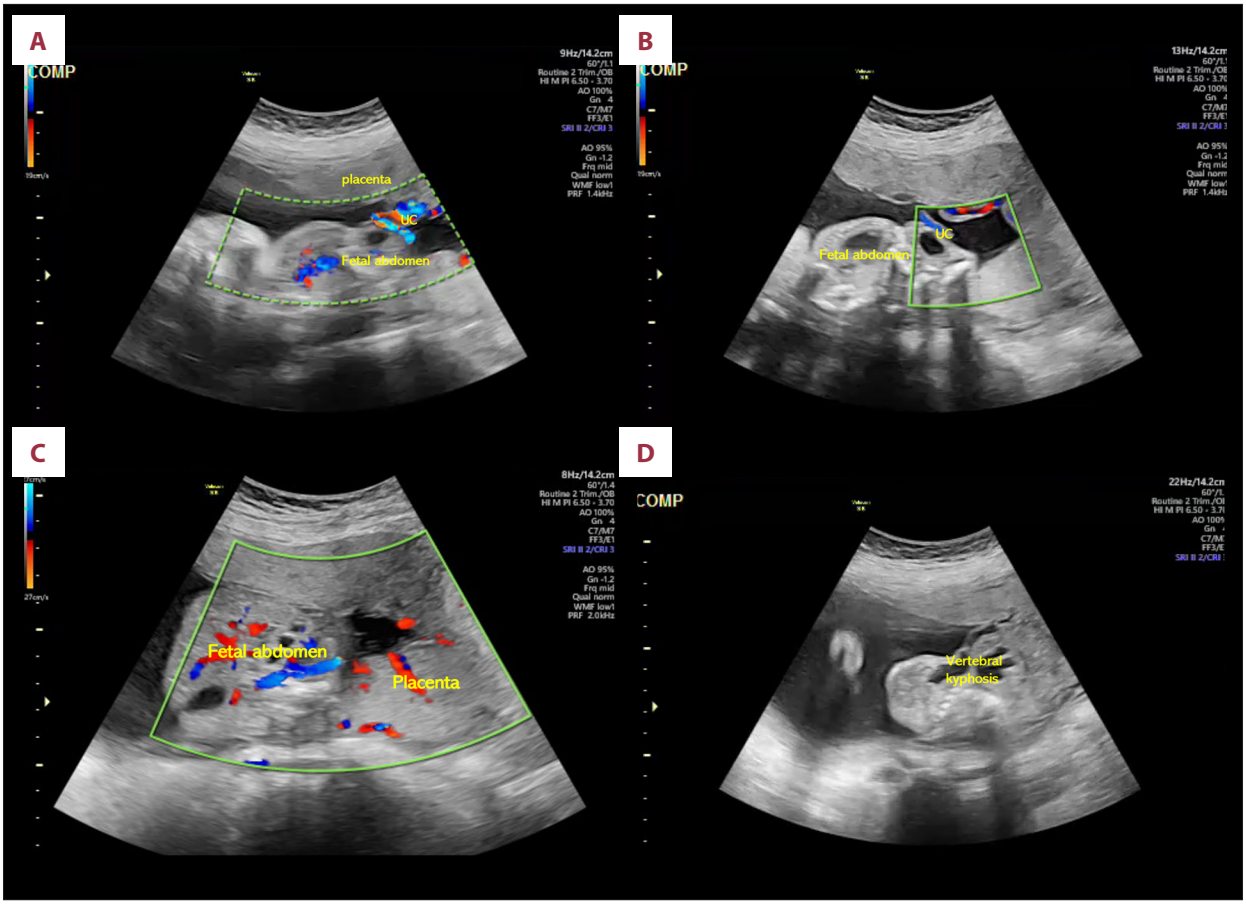
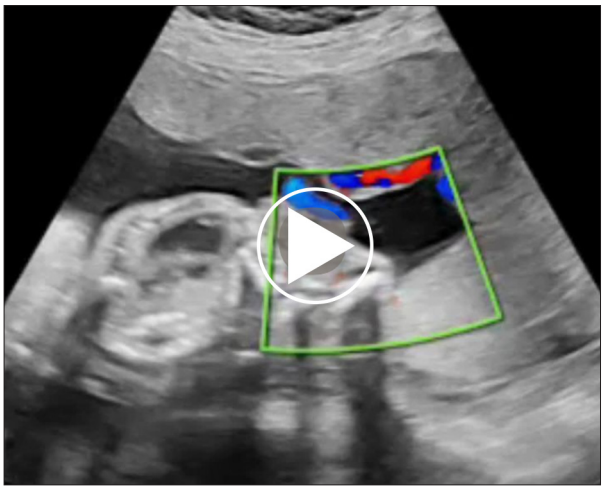


Figure 2. Fetomaternal ultrasound result revealing body stalk anomaly: (**A, B**) very short umbilical cord (UC); (**C**) severe abdominal wall and limb defect; and (**D**) severe limb defect.

poor fetal prognosis and the need to prevent further vascular growth, termination of pregnancy was recommended. During surgery, significant bleeding from the placental bed and cervix was encountered, and despite attempts to control the hemorrhage through figure-8 sutures and compression, the bleeding persisted due to uterine atony. As a result, a hysterectomy was ultimately necessary to manage the ongoing postpartum hemorrhage. This case highlights the challenges of performing major surgery in the presence of a lethal fetal anomaly, in which maternal safety must be prioritized.

Case Report

A G2P1A0 29-year-old woman came to the Maternal-Fetal Medicine Unit for prenatal diagnosis at 29 weeks of gestation. The patient was asymptomatic and came with a referral diagnosis of suspected omphalocele. Her obstetrical history revealed a previous normal pregnancy that ended with a cesarean section, due to a contracted pelvis. She had never undergone an ultrasound sonography examination during the first trimester.



Video 1. Ultrasound video showing a very short umbilical cord.

At the referral hospital, obstetric ultrasound was performed, with the result of a singleton breech pregnancy equal to 27 to 28 weeks of pregnancy (**Figure 1**) with a severe abdominal wall defect, consisting of the intestine, liver, and stomach (**Figure 2**), severe kyphoscoliosis (**Figure 2D**), and a very

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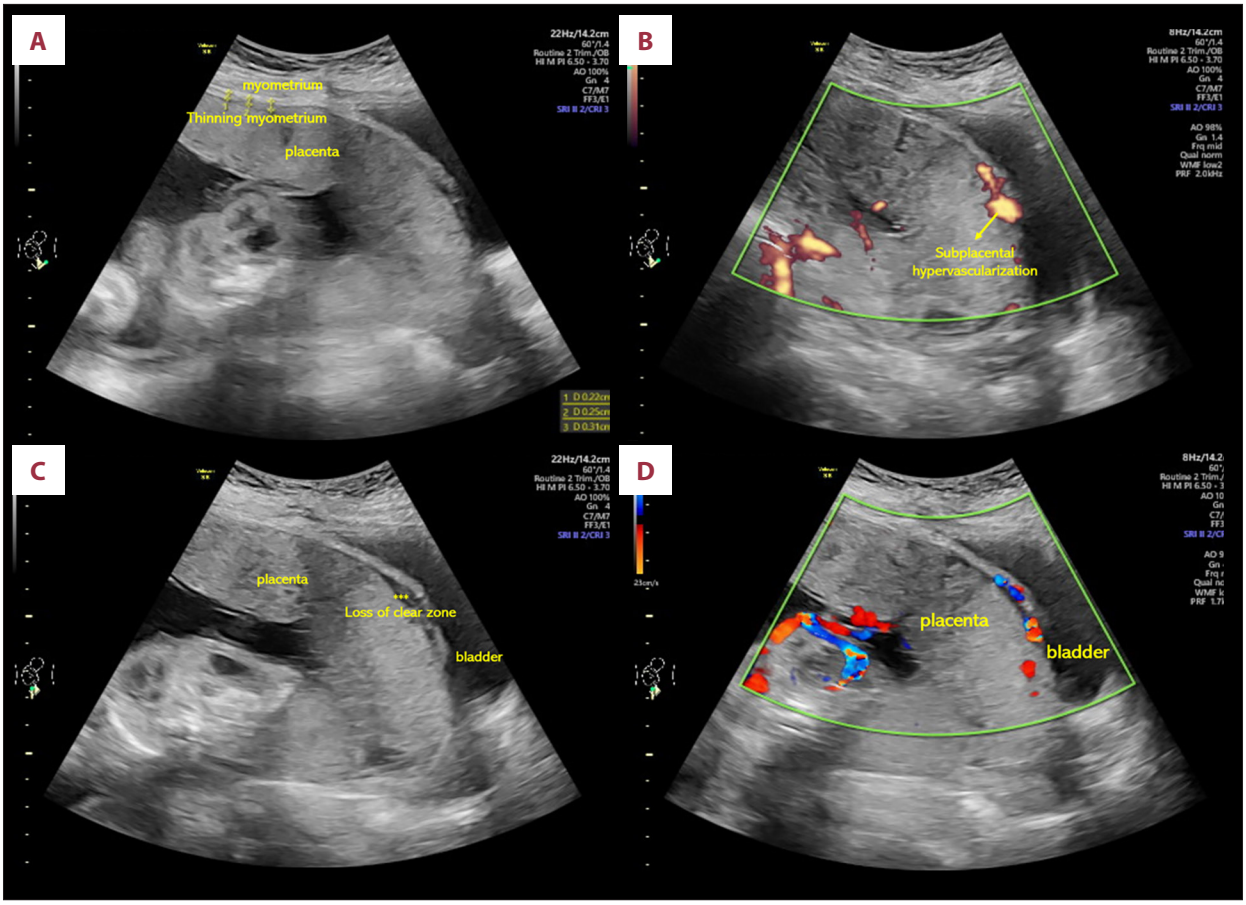

Figure 3. Ultrasound scan result showing features of placenta accreta spectrum: (**A**) thinning of the myometrium; (**B**) suplacenta hypervascularization; (**C**) loss of clear zone; and (**D**) subplacental hypervascularization on Doppler view.

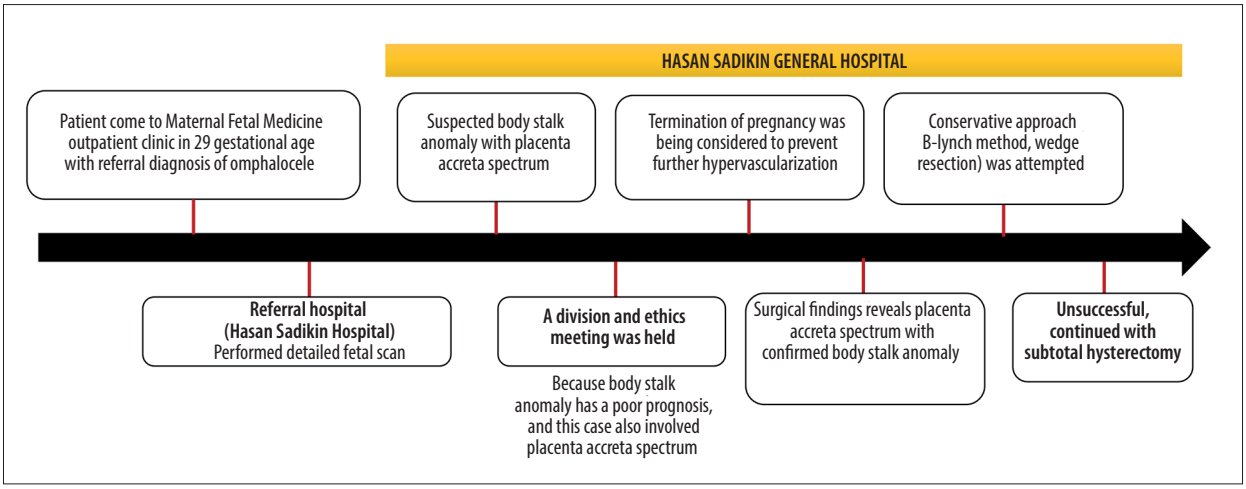

Figure 4. Timeline of patient's case management.

rudimentary umbilical cord (**Video 1**) with limb defect. Based on ultrasound findings, the patient probably had a body stalk anomaly, with differential diagnosis of pentalogy of Cantrell or gastroschisis. Ultrasound examination of the placenta revealed loss of retroplacental-myometrial radiolucent interface and increased vascularity at the placental bed (**Figure 3**). The placenta location in the anterior corpus extending toward the internal uterine ostium with loss of clear zone and subplacental hypervascularization clarified this patient had total placenta previa with placenta accreta spectrum.

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Figure 5. Outcome result of the fetus. The fetus exhibits an extremely short umbilical cord measuring approximately 15 mm in length. (**A**) Illustrates a frontal view highlighting vertebral kyphosis; (**B**) lateral view of the fetus; and (**C**) closer view of the abdominal region, displaying the herniated abdominal contents.

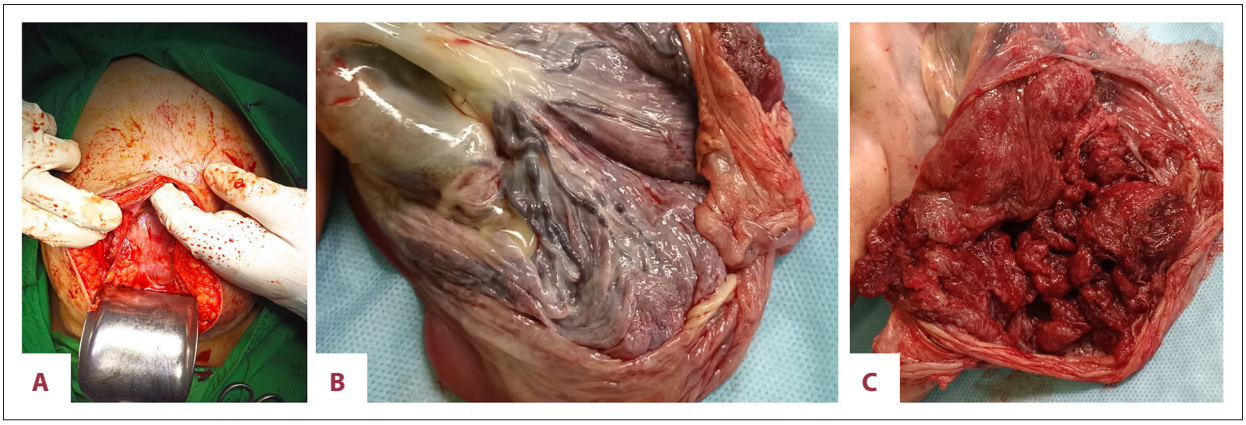

Figure 6. Intraoperative and pathological findings in placenta accreta spectrum with subtotal hysterectomy. (**A**) Initial intraoperative findings show adhesions between the anterior uterine wall and omentum. Adhesiolysis was performed to free the structures. The incision was made transperitoneally approximately 2 cm above the serosa-bladder reflection to avoid the placenta insertion. (**B, C**) Gross view of the placenta showing the placenta still in situ. No bridging vessels or signs of hypervascularization were observed.

Laboratory test results during admission were within reference ranges, without any sign of anemia. The patient then received a diagnosis of suspected body stalk anomaly, with placenta accreta spectrum. Since poor prognosis of the baby and further gestation would increase hypervascularization, the patient chose to terminate the pregnancy. The placenta accreta spectrum necessitated a collaborative meeting between the ethics committee and clinicians from several fields. The specialists reached an aggreement to terminate the pregnancy via cesarean delivery, due to the poor prognosis of the fetus and the fact that prolongation of the pregnancy would exacerbate the degree of hypervascularization. **Figure 4** shows the timeline of case management.

A cesarean delivery was performed, with the indication of placenta accreta spectrum at 30 weeks of pregnancy, to facilitate conservative management. The surgical procedure involved careful steps to address placental attachment and manage complications. After performing antiseptic measures, a median inferior incision was extended, as needed. Upon opening the peritoneum, adhesions were observed between the anterior uterus and omentum, which were carefully lysed. Exploration of the uterine surface showed no hypervascularization, bridging vessels, or bulging. The vesicouterine fold and bladder boundaries were identified.

Preoperative ultrasound was used to locate the placenta and guide the incision site, ensuring avoidance of the placental attachment site. The baby was delivered by gently luxating the head. After the incision, the baby was delivered with a body weight of 800 g, body length of 25 cm, and an APGAR score of 1-1. The baby exhibited a very short umbilical cord and severe abdominal wall and limb defects, consistent with body

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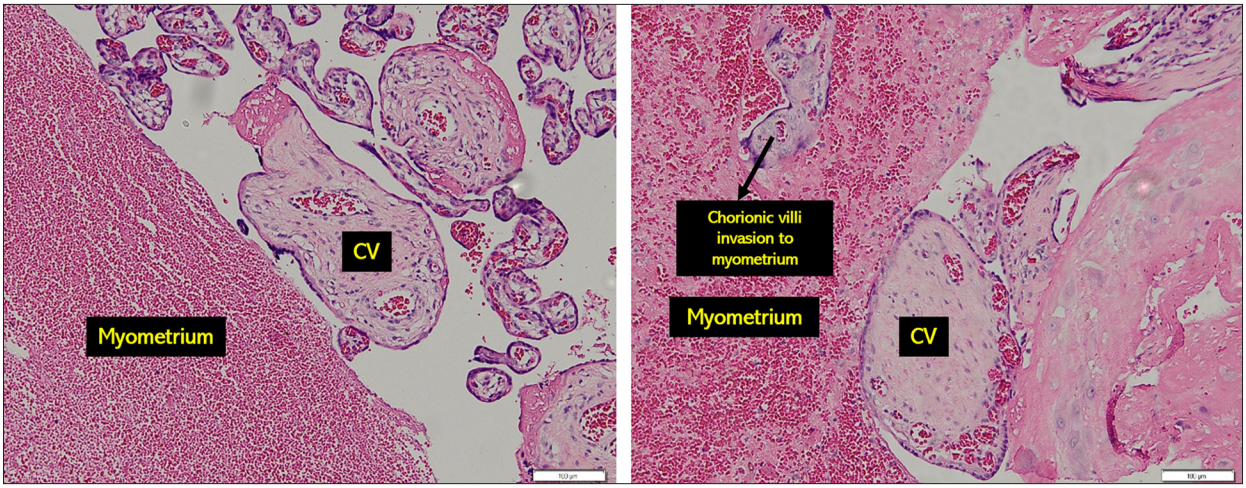

Figure 7. Histopathology specimen of chorionic villi (CV) structure and myometrium. Note the black arrow shows CV invasion to the myometrium.

stalk anomaly. Given the poor prognosis, further resuscitation was not performed. **Figure 5A-5C** shows the outcome of the fetus. The length of the umbilical cord was roughly 15 mm, indicating an extremely short umbilical cord. The vesicouterine fold was successfully separated and retracted caudally. Double ligation of the right and left uterine arteries, as well as the vesical arteries, was performed, followed by a figure-8 suture to control bleeding from the upper vaginal area. The placenta was delivered manually, with the operator carefully using the back of the hand to peel it away from the anterior uterine wall while gently freeing the placental membranes using ovum forceps. The placenta weighed 250 g and measured 15×15×1.5 cm (**Figure 6A-6C**).

Despite these efforts, active bleeding from the cervix was noted. A large gauze (Big Hass) tampon was inserted, and a Foley catheter was used as a tourniquet to manage bleeding. Wedge resection was performed, with size 4×3×1 cm.

Due to continued hemorrhage from the placental bed, further bleeding control was attempted with figure-8 sutures, using chromic No. 2 sutures. However, active bleeding from the cervix persisted, and hemostasis was difficult to reach. Compression tests failed to stop the bleeding, and uterine contractions remained inadequate, due to uterine atony. Given the ongoing hemorrhage, a subtotal hysterectomy was performed to control the bleeding and ensure maternal safety. Histopathological examination confirmed the diagnosis of placenta accreta spectrum, with chorionic villi invading the myometrium (**Figure 7**).

Discussion

This case highlights the challenges of managing a rare and severe condition, body stalk anomaly, complicated by placenta accreta spectrum. It emphasizes the importance of early prenatal screening, advanced imaging for diagnosis, and a multidisciplinary approach to high-risk pregnancies.

A body stalk anomaly is an uncommon and sporadic abnormality of the abdominal wall, occurring in a reported range of frequencies from approximately 1 in 7500 pregnancies to 0.32 in 10 000 births [10], while another source states 1 in 14 000 to 22 000 pregnancies [4]. This abnormality is thought to be the consequence of abnormalities in the development of cephalic, caudal, and lateral embryonic folding and a defect in the closure of the body wall during embryogenesis [4,10]. Other literature states that it is a severe abdominal wall defect in which the peritoneal cavity is exposed to the extra-embryonic coelom, with variation from a very short to an absent umbilical cord that attaches the fetus' abdominal wall to the placenta [11,12].

The differentiation between body stalk anomalies and other abdominal wall defects is attributed to the presence of the liver and intestine within the extra-embryonic coelom. This disorder should be considered when prenatal ultrasonography identifies a significant abdominal defect, with the existence of the liver and intestine in the extra-embryonic coelom. Additionally, the presence of vertebral deformities, such as kyphosis or scoliosis, along with a short or nonexistent umbilical cord, can further support the suspicion of this illness. Karyotyping is not recommended in cases of body stalk anomalies, due to the absence of any known association with chromosomal abnormalities [4,10-12].

Three theories possibly explain this kind of anomaly. Potential etiologies of body stalk anomaly include early amniotic rupture accompanied by direct mechanical compression and the presence of amniotic bands, which can account for the observed

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limb amputations. Additionally, vascular disruption during the early stages of embryonic development or abnormalities in the germinal disk could explain the occurrence of not only vertebral anomalies but also severe abdominal wall abnormalities [13,14]. The prevailing hypothesis suggests that the rupture of the amnion occurs prior to the closure of the coelom. This rupture, which can lead to deformities in the abdomen, spine, and limbs, occurs when the lower fetus enters the extra-embryonic coelom, resulting in a severe amniotic band sequence [11,15].

The germinal disk abnormality may imply a complete body fold inability along the cephalic, caudal, and lateral axes. The common physiological mechanism of body folding divides the intra-embryonic coelom, which will become the peritoneal cavity, from the extra-embryonic. The body stalk and umbilical cord are also created during this step. Abnormal head folding causes thoracic wall and epigastrium deformity, enabling ectopia cordis. A broad-base amnio-peritoneal sac forms when midabdominal contents protrude due to improper lateral folding. This sac is linked to the placental chorionic plate at the periphery by a short umbilical cord or without one [11,12].

The development of severe scoliosis and anomalies in the axial skeleton occurs as a consequence of the asymmetrical development of the spine and thoracic cavity, caused by the extrusion of intra-abdominal contents. The occurrence of malrotation in the spine and insufficient closure of the pelvis can result in the manifestation of malrotated limbs and/or club foot [11,12]. In our patient, severe abdominal wall defect was found (consisting of intestines, liver, and stomach), with a very short umbilical cord, limb defect, and severe kyphoscoliosis. There were no specific risk factors in this patient.

The differential diagnosis of abdominal wall abnormalities includes exomphalos, gastroschisis, omphalocele-exstrophy-imperforate anus-spinal defects complex, and the pentalogy of Cantrell. Since the patient presented during the second trimester, it was possible to observe pronounced abdominal wall deformities that are connected to the placenta, as well as severe kyphoscoliosis and the lack of a freely floating umbilical cord. These findings were indicative of a diagnosis of bladder exstrophy-epispadias complex. Exomphalos is characterized by the presence of herniated viscera near the base of the umbilical cord, with a visible free-floating chord within the amniotic cavity. Deformation and disruption of several structures, such as craniofacial structures and limbs, might be observed in cases of early amnion rupture. The disruption of amniotic membrane continuity characterizes abdominoschisis resulting from amniotic bands, while the umbilical cord remains unattached [16,17].

The diagnosis and management of a body stalk anomaly present difficulties because of its intricate appearance and the elevated risk of fetal morbidity and mortality. Prenatal screening and ultrasonography are essential for early detection, organization of suitable prenatal care, and delivery management [4]. Placenta accreta spectrum represents a considerable global health issue, especially given the rising incidence of cesarean deliveries globally. The incidence of placenta accreta spectrum is increasing in conjunction with the frequency of cesarean deliveries. This syndrome presents significant risks, including severe hemorrhage and the necessity for hysterectomy, resulting in heightened maternal morbidity and mortality. The timely identification of placenta accreta spectrum, particularly during the first trimester, is essential for enhancing results and safeguarding uterine function, whenever feasible [18]. The global healthcare community continues to emphasize the importance of early diagnosis and multidisciplinary management approaches to optimize care for affected women.

Unfortunately, our patient presented with placenta accreta spectrum. Placenta accreta would result in a significant restructuring of the arcuate and radial arteries, a deficient activation of certain cell receptors, and an increased secretion of trophoblast-modulating molecules, such as vascular endothelial growth factor, as a result of low oxygen levels. Consequently, hypoxia would promote the growth of trophoblast cells, while normoxia would hinder their growth. Without the presence of decidua in the myometrium, the trophoblastic invasion intensifies, as the inhibitory effects mediated by the metalloproteinases in the extracellular matrix of the decidua are absent [19]. To date, there is no report linking body stalk anomaly and placenta accreta spectrum. However, it is plausible that the co-occurrence of body stalk anomaly and placenta accreta spectrum may be due to its rare incidence. Currently, the veracity of this correlation remains uncertain, as there is a lack of published studies investigating this matter. Alternatively, the occurrence of body stalk abnormality and placenta accreta spectrum may be more likely, due to the widespread use of cesarean procedures. A study conducted by Okido et al suggests there may be a link between the limb body wall complex abnormality and hypoxia diseases, resulting from insufficient placental development in previous uterine scarring [20].

The term “placenta accreta spectrum” is commonly used to encompass the 3 conditions of accreta, increta, and percreta [21-23]. Placenta accreta spectrum can sometimes be confused with cesarean scar dehiscence, as both can present similar findings on imaging and laparotomy. However, in cesarean scar dehiscence, the placenta separates at birth, leaving a thin, distended lower uterine segment at the site of the prior scar. In contrast, placenta accreta spectrum involves abnormal adherence of the placenta to the myometrium, which leads to difficulty or failure in placental detachment, resulting in severe postpartum hemorrhage and potential hysterectomy [24].

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Placenta accreta is not associated with placental hypoxia or fetal malformations. Instead, the primary issue in placenta accreta spectrum is linked to hypervascularization and peri-isthmic hyperemia, which can severely complicate attempts at uterine conservation if extreme fibrosis or insufficient dissection is present. Furthermore, recent literature has highlighted the potential for early detection of placenta accreta spectrum, particularly in the first trimester, in cases of placental implantation in cesarean scar tissue (commonly referred to as "scar pregnancy" or "baby accreta"). Early identification of this condition increases the likelihood of preserving the uterus through early intervention [25].

A recent study conducted by Palacios-Jaraquemada et al shown that lower placenta accreta spectrum parametrial involvement is infrequent but correlates with increased maternal morbidity. Upper and lower parametrial placenta invasion provide distinct surgical risks and necessitate varied technical techniques; therefore, precise diagnosis is essential.Women with a clinical history of manual placental removal, abortion, or recurrent dilation and curettage must be meticulously evaluated to determine a potential placenta previa incidence. For patients with high-risk histories or inconclusive ultrasound findings, a T2-weighted MRI is advised [26]. However, in the present case, no MRI was performed before the intervention. This decision was influenced by the acute nature of the patient's condition and the poor fetal prognosis, due to body stalk anomaly. Given the urgency of managing both the placental attachment and the lethal fetal condition, the team prioritized immediate surgical intervention. While MRI could have provided more detailed information about placental invasion, the decision to proceed without it was made based on the clinical presentation and the need to avoid further delays in managing the patient's high-risk situation [9].

Imaging played a crucial role, with ultrasound findings, such as loss of the retroplacental-myometrial interface and hypervascularity, helping to identify placenta accreta spectrum. In placenta accreta spectrum, the absence or defective development of the decidua basalis and the Nitabuch layer contributes to abnormal placental attachment. This leads to substantial maternal morbidity and sometimes necessitates cesarean hysterectomy to manage the severe hemorrhage that often accompanies placenta accreta spectrum [27]

Ultrasound findings on this patient that match the diagnosis of placenta accreta were as follows [28,29]: (1) thinning of retroplacental myometrium (**Figure 3A**); (2) irregularity or disruption of the echogenic interface between the maternal bladder and uterine serosa (**Figure 3B**); (3) loss of clear zone (**Figure 3C**); and (4) increased vascularity of the bladder-serosal interface, with a bridging vessel (**Figure 3D**).

Ultrasound findings in this case showed a match with the diagnosis of placenta accreta spectrum (**Figure 3A-3D**). Our patient already received a diagnosis of placenta accreta spectrum with lethal congenital anomaly; therefore, she chose to terminate the pregnancy. The decision-making process in this exceptional case was undoubtedly a challenging situation, given the unfavorable prognosis for the fetus and the potential irreversibility of the patient's nonviable uterus, which would render her unable to conceive during her reproductive years. The Society for Maternal-Fetal Medicine advises scheduling a cesarean delivery, along with the possibility of a hysterectomy, during the late preterm period, specifically between 34 0/7 and 35 6/7 weeks of gestation, for cases in which placenta accreta spectrum is suspected prenatally [30]. The patient in this case had ultrasound findings consistent with placenta accreta spectrum, resulting in the inability to perform a vaginal birth.

Cesarean hysterectomy is regarded as the optimal approach for addressing invasive placentation. Nonetheless, this extreme method is linked to significant rates (40%-50%) of severe maternal complications, primarily due to hemorrhage and damage to adjacent organs during the procedure, with mortality rates reaching up to 7% as a result of massive, untreatable hemorrhage [31]. Significant hemorrhaging can arise from the spontaneous separation of a portion of the placenta or through manual separation of the placenta. Hemostasis is achieved through a range of procedures, such as gauze tamponade, suturing of the bleeding site (including Z suture, U-shaped whole myometrial suture, interrupted circular suture), compression suture, and arterial ligation. If bleeding remains unmanageable, a total hysterectomy is indicated. Management decisions upon clinical diagnosis of placenta accreta typically necessitate hysterectomy in instances when FIGO grade exceeds II [32].

Conservative management refers to the approach taken to treat both an abnormally adherent (placenta acreta) and invasive placenta (placenta increta and percreta). The goal of this approach is to prevent the need for peripartum hysterectomy and the associated morbidity and consequences. In 2 scenarios, a conservative management can be considered: (1) when intraoperative findings suggest that hysterectomy is likely to be complicated and carries a significant risk of massive hemorrhage or injury to adjacent tissues, which may be mitigated by leaving the placenta in situ; and (2) for women who wish to have children in the future or whose fertility is closely tied to their social status and self-esteem [31]

The international literature has described 4 distinct primary techniques of conservative management. (1) The extirpative technique involves manually removing the placenta. (2) The expectant approach involves keeping the placenta in place. (3) One-step conservative surgery involves removing only the area affected by the accreta. (4) The triple-P operation involves suturing around the accreta area after it has been removed [33]

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Prior to terminating the pregnancy, it is advisable to obtain the patient's consent of the potential need for surgical extension, possibly leading to a hysterectomy. Our patient expressed a desire for future fertility, and she was counseled about the possibility of requiring hysterectomy in the event of complications. Ultimately, due to uncontrollable postpartum hemorrhage, a hysterectomy was performed. Histopathological examination confirmed the diagnosis of placenta accreta, with chorionic villi invading the myometrium (**Figure 4**). Histopathology findings confirmed the presence of placenta accreta spectrum in the myometrium, which was shown by the invasion of the chorionic villi to the myometrium. Conducting thorough surgical staging in placenta accreta spectrum facilitates the prompt detection of parametrial placenta invasion prior to using any dissection techniques that can result in unforeseen substantial hemorrhaging. Understanding what actions to take or avoid is crucial for preventing needless organ ablation and unmanageable hemorrhaging [26].

In the present case, the decision to terminate the pregnancy was made after considering both the poor fetal prognosis and the increasing maternal risks. Despite conservative efforts, persistent hemorrhage necessitated a subtotal hysterectomy to ensure maternal safety. This case also highlights the importance of a coordinated, multidisciplinary approach, as collaboration among specialists was essential for optimizing care in this complex scenario. Ultimately, it provides valuable insights into clinical decision-making and management strategies for similar high-risk pregnancies.

Conclusions

Body stalk anomaly is an exceedingly rare condition, and its combination with placenta accreta spectrum, as seen in this patient, results in a very poor prognosis. Given the complex nature of this case, earlier detection of both conditions, particularly through first-trimester screening and preemptive management of abnormal placentation, might have offered an opportunity to explore alternative treatment options that could have better preserved the patient's fertility. However, once severe postpartum hemorrhage occurred, cesarean hysterectomy was necessary to prevent further maternal complications. Since the patient had already undergone a cesarean hysterectomy, preconceptional counseling was no longer relevant. However, future counseling should focus on improving the patient's quality of life after surgery, addressing physical recovery, mental health, and long-term well-being.

Informed Consent

The patient received a comprehensive explanation regarding the case's particulars and the images to be included in the case report. The patient provided consent of the case report.

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Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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