CASE REPORT

Adenomyosis in pregnancy mimicking morbidly adherent placenta

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SUMMARY

The objective of this study was to illustrate a false-positive diagnosis of adherent placenta due to underlying adenomyosis. A 34-year-old woman was diagnosed for placenta previa totalis with adherent placenta at 33 weeks, based on the findings of loss of clear space or distinguishing outline separating the placenta and uterine wall, presence of intraplacental lacunae and densely atypical vessels in the lesion. Caesarean hysterectomy was performed at 35 weeks. Pathological findings revealed placenta previa totalis with adenomyosis beneath the placenta at the lower segment, with no adherent placenta. In conclusion, this report underlines the importance of possible false-positive test of prenatal ultrasound and MRI findings of adherent placenta caused by underlying adenomyosis which could obliterate the outline distinguishing the placenta and myometrium and atypical vessels secondary to decidualisation and hypervascularity from pregnancy. This case may probably encourage physician to beware of false-positive test of adherent placenta due to adenomyosis.

BACKGROUND

Morbidly adherent placenta is the placenta unusually adhered to the implantation site, associated with absent or scanty decidua and the physiological line of cleavage. MAP could be categorised into three subgroups: (1) placenta accreta (chorionic villi attach to myometrium rather than decidua), (2) placenta increta (chorionic villi penetrate into the myometrium) and (3) placenta percreta (chorionic villi penetrate through the myometrium to the uterine serosa or adjacent organs). The disorder is known to be associated with significant maternal morbidity/mortality and often presents a significant obstetric challenge, at times resulting in life-threatening haemorrhage and the need for peripartum emergency hysterectomy.¹ The prevalence of the morbidly adherent placenta is markedly increased in cases of placenta previa 9.3%, and 0.04% of women with and without placenta previa,² and previous uterine surgery especially caesarean section. Currently, prevalence of adherent placenta is rising because of an increasing rate of caesarean deliveries,³ a risk factor of placenta previa as well as adherent placenta. In addition, advanced maternal age, multiparity and previous curettage are also significant risk factors. It has been well accepted that prenatal diagnosis of adherent placenta is feasible and helpful to reduce the risk of maternal morbidity/mortality.^{4 5} The accurate diagnosis of morbidly adherent placenta enables us to be well prepared for severe postpartum haemorrhage or elective delivery, leading to avoidance of severe morbidity

secondary to massive haemorrhage. For example, Robinson and Grobman⁴ showed that elective delivery for pregnancies with placenta previa and adherent placenta at 34 weeks of gestation were associated with the highest quality-adjusted life years under a variety of circumstances. Nevertheless, diagnostic accuracy is not perfect, and a false-positive test could result in disturbance to routine clinical services and wastage of human and economic resources. In relatively large studies, the sensitivity and specificity of ultrasound for prediction of placenta accreta are 77–90% and 71–98%, respectively.^{6–9} Sonographic findings suggestive of placenta accreta include the following^{6 8 10}: (1) loss of placental homogeneity, which is replaced by intraplacental sonolucent lacunae or vascular lake with turbulent flow, (2) loss or thinning of the normal hypoechoic area behind the placenta, (3) disruption of the normally continuous white line representing the bladder wall–uterine serosa interface and (4) hypervascularity of serosa–bladder interface. The objective of this report was to illustrate the false-positive case of adherent placenta because of underlying adenomyosis confined to the lower uterine segment at the area beneath placental attachment, which has never been described elsewhere. The present case may probably encourage physicians to beware of false-positive test of adherent placenta due to adenomyosis, even in case of typical imaging.

CASE PRESENTATION

A 34-year-old woman, G2 P1001, was referred to our hospital at 33 weeks of gestation because of antepartum haemorrhage. She had had a history of uterine curettage due to abnormal bleeding suspected of incomplete abortion. She had attended antenatal care clinic at a primary healthcare hospital with uneventful pregnancy course and no serious medical or familial diseases. She was diagnosed for high-grade squamous cell intraepithelial lesion of the cervix in early gestation with a plan for definite treatment after birth. Fetal scan at mid-pregnancy showed no abnormality. At 33 weeks of gestation, she had two episodes of haematuria. Physical examination on admission revealed unremarkable and uterine size compatible with gestational age. Transabdominal and transvaginal ultrasound examinations revealed a structurally normal fetus with normal biometry with placenta previa totalis. The placenta was mostly attached to the anterior wall of the uterus including the lower segment and covered the internal os with extension to some portion of the posterior wall.

Grey-scale ultrasound could show clear space or echolucent area beneath the placenta with clear outline distinguishing the uterine wall from the

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Figure 1 Mid-sagittal scan of transabdominal ultrasound showing normal placental attachment in the upper uterine segment and loss of interface outline (arrowhead) in the lower segment in which placental texture and uterine wall cannot be distinguished and several lacunae are demonstrated.

placenta in the upper uterine segment (figure 1). However, the separating outline abruptly ended at the lower uterine segment and from this point below the placenta and uterine wall was no more distinguishable. In the lower segment, loss of echolucent area or clear space between the placenta and the uterine wall and the presence of several lacunae with turbulent high-velocity flow in the mixed area were demonstrated. In addition, hypervascularity at the serosa–bladder wall interface with densely confluent anarchic vessels that are much more prominent than that in the upper uterine segment was also clearly visualised on colour flow mapping (figure 2 and video 1).

MRI findings were consistent with the ultrasound findings (figure 3). Placenta previa covered most area of the anterior lower segment and the entire internal os. Lower part of the placenta and the anterior wall of the lower uterine segment could not be clearly distinguished from each other. The bladder wall seems intact, without gross involvement and absence of a protruding mass into the bladder. Prenatal diagnosis of placenta previa totalis with morbidly adherent placenta was made with suspected placenta percreta and bladder involvement, suggesting clinical haematuria.

Threatened preterm labour with minimal bleeding per vagina was observed on the day of admission. However, spontaneous remission occurred without tocolysis. The patient was admitted and monitored for bleeding and fetal well-being. No serious

Figure 2 Mid-sagittal scan of transabdominal ultrasound of colour flow mapping showing several lacunae in the lower segment and hypervascularity.

Video 1 Video clip of transvaginal ultrasound examination with color flow mapping: Sagittal scan of the lower segment shows hypervascularity with several lacunae and no distinguishing outline between the placenta and uterine wall.

bleeding was observed during admission. Spontaneous labour occurred at 35 weeks of gestation. Before caesarean section, cystoscopy by a urologist revealed congestion of the vessels at the posterior wall, presumably caused by interstitial cystitis, but intact bladder mucosa. Classical caesarean section was performed, giving birth to a male child weighing 3120 g, with Apgar score 9 at 5 min. Relied on preoperative imaging diagnosis, together with the presence of high-grade squamous cell intraepithelial lesion of the cervix, total hysterectomy with intact placenta was performed without trial of manual placental removal. Endometriosis and adhesions between bladder wall and anterior uterine segment were observed and one endometrioma was accidentally ruptured with chocolate content spillage during the operation.

Pathological findings revealed placenta previa totalis with multiple lesions of adenomyosis, as seen in the preoperative images, with no placenta adherens (figure 4). The placenta could be manually removed from the uterine wall with no force. The microscopic finding confirmed no placental adherens, without trophoblastic villi invasion to myometrium (figure 5). However, multiple lesions of adenomyosis with marked stromal decidualisation, intervening myometrial bundles of the uterine wall beneath the placental attachment at the lower segment, were demonstrated (figure 6). In addition, multiple endometriosis were located along the outer surface of the lower uterine segment juxtaposed to the bladder wall (figure 7).

OUTCOME AND FOLLOW-UP

The patient was healthy without any serious sequelae on the follow-up visit at 6 weeks after operation.

DISCUSSION

In the present case, clinical manifestation and imaging of MRI and ultrasound were strongly suggestive of morbidly adherent placenta, as described several times in the literature.^{11–17} Moreover, placenta previa in this case placed the patient to be at a very high risk of placental adherens, as already known that placenta previa is an important factor of morbidly adherent placenta (OR=51.42).¹⁸ Furthermore, this case also had a previous history of questionable uterine curettage because of criminal abortion, another risk factor for placenta adherens. Prenatal diagnosis of morbidly adherent placenta in the present case was based on the findings as follows: (1) loss/irregularity of echolucent area between placenta demonstrated by ultrasound and MRI, (2) the presence of intraplacental lacunae using grey-scale

Figure 3 (A) Mid-sagittal plane of MRI showing normal placental attachment in the upper uterine segment and loss of interface outline in the lower segment in which placental texture and uterine wall cannot be distinguished; (B) overlay labels of the image in (A).

and colour Doppler imaging, (3) hypervascularity at the serosa-bladder wall interface with densely confluent anarchic vessels that seemed to protrude into the bladder lumen. However, our case indicated that such findings could be secondary to the causes other than the adherent placenta. Adenomyosis in the uterine wall together with deep endometriosis located between bladder wall and uterine wall could have the same appearance as adherent placenta. How the lesions give the findings mimicking adherent placenta is not clear. Nevertheless, it might be possible that the pressure effect from adenomyosis could obliterate the clear space or echolucent area between the placenta and uterine wall and obscure the placental outline. Furthermore, adenomyosis might be more highly vascularised during pregnancy, resulting in more complex and tortuous vessels. In addition, endometriotic cysts between the bladder wall and lower uterine segment could simulate lacunae as seen in adherent placenta.

It is well accepted that prenatal diagnosis of adherent placenta is feasible and helpful. Certainly, a high sensitivity rate is desirable to facilitate the preparation and management team, either obstetricians or anaesthesiologists. Nevertheless, diagnostic

accuracy is not perfect, either using transvaginal high-resolution ultrasound or MRI, and a false-positive test could result in disturbance to routine clinical services and wastage of human and economic resources or over-treatment like the present case. The lesson from this case is that even in cases with typical imaging suggestive of adherent placenta, intraoperative trial of manual placental separation with extremely high precaution for definite diagnosis may be encouraged with a well preparedness for excessive bleeding.

Other than false-positive impression of adherent placenta, the pitfall in management of the present case might include a jump to total hysterectomy without trial of manual separation with precaution. Our case highlights the importance of well-documented pre-existing lesions of the uterus, including adenomyosis as in this case to avoid confusion with subsequent adherent placenta. One should remember that typical characteristics of adenomyosis may become atypical or bizarre during pregnancy secondary to decidualisation caused by hormonal effects or increased vascularisation especially when adenomyosis is confined to placental area.¹⁹ It was possible that in our case the decidualised adenomyosis might result in atypical characteristics

Figure 4 Gross section of the lower uterine segment showing lacunae and adenomyosis beneath placental attachment and endometriosis at the outer surface.

Figure 5 Microscopic appearance of the lower uterine segment showing placental villi attached to the decidua (fibrinoid layer) not reaching myometrium.

Figure 6 Microscopic appearance of the uterine muscle at the lower segment beneath placental site showing a marked decidualisation of adenomyosis localised between muscle bundles (D, decidualisation of adenomyosis; M, myometrium).

of ultrasound and MRI appearances of adenomyosis. Note that uterine-placental outline ended abruptly at the lower uterine segment, corresponding to the pathologically reported adenomyosis. Placental textures and the underlying adenomyosis were mixed with obscure outline. Moreover, during pregnancy, the blood volume in uterus has been markedly increased. Therefore, the adenomyosis beneath the placental attachment may possibly cause hypervascularity, leading to more enlarged vessels than seen in other area of the placenta and several lacunae in the area. Together with deep endometriosis between bladder wall and uterine wall, the imaging seemed to be suggestive of placental invasion to the entire thickness of the lower uterine segment.

Adenomyosis is a common disease, characterised with the presence of endometrial tissue, glands and stroma, within the myometrium, below the endomyometrial junction. In non-pregnant state, ultrasound findings suggestive of adenomyosis include subendometrial linear striations, a regularly enlarged uterus with a globular appearance and myometrial cysts and heterogeneous myometrium.²⁰ Likewise, on MRI, adenomyosis is typically visualised as either diffuse or focal thickening of the junctional zone forming an ill-defined area of low signal intensity, sometimes with embedded bright foci on T2-weighted

Figure 7 Microscopic appearance of the uterine outer surface at the lower segment juxtaposed to the posterior bladder wall showing a marked decidualisation of endometriosis.

image.¹⁹ The ectopic endometrium of adenomyosis is relatively inactive or proliferative, but during pregnancy, functional changes and stromal decidual reactions in adenomyosis may be encountered, histologically characterised by remarkable stromal oedema and enlarged stromal cells to form polygonal or round decidual cells. This is due to the fact that adenomyosis is an oestrogen-progesterone-dependent disorder, as it is associated with persistently elevated oestrogen levels.²¹

In conclusion, the present case underlines the importance of possible false-positive test of prenatal ultrasound and MRI findings of morbidly adherent placenta caused by underlying adenomyosis and deep endometriosis in the lower uterine segments, which could obliterate the outline distinguishing the placenta and myometrium and atypical vessels secondary to decidualisation and hypervascularity from pregnancy.

Learning points

- ▶ Typical ultrasound findings suggestive of adherent placenta can be caused by adenomyosis beneath the placenta.
- ▶ Ultrasound findings of adenomyosis during pregnancy can be atypical because of decidualisation and hypervascularity.
- ▶ Even in cases with typical imaging suggestive of adherent placenta, intraoperative trial of manual placental separation with extremely high precaution for definite diagnosis may be encouraged with a well preparedness for excessive bleeding.

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