

Retained Placenta Variants: Adhered Placenta to a Large Non-Pedunculated Submucosal Uterine Myoma Following a Vaginal Delivery at Ekona Subdivisional Hospital, a Case Report and Literature Review

William Ntchompbopughu Tih* and Paul Mbanyi Mbanyi

Department of Obstetrics and Gynaecology, Faculty of Health Sciences, University of Buea, Ekona Subdivisional Hospital, Cameroon.

*Correspondence:

William Ntchompbopughu Tih, Department of Obstetrics and Gynaecology, Faculty of Health Sciences, University of Buea, Ekona Subdivisional Hospital, Cameroon.
ORCID: 0000-0002-8306-9788.

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ABSTRACT

We are reporting the case of a retained placenta adhered and implanted to a large non-pedunculated submucosal myoma in a primary healthcare facility in the Southwest region of Cameroon. This is the case of a 31-year-old G1P0 patient, blood group A rhesus positive at 39 3/7 weeks' gestation whose labor progressed to the vaginal delivery of a life male baby with 1st and 5th minute APGAR score of 8/10 and 10/10. A single ultrasound was done in the third trimester (36 weeks) of her pregnancy and she attended only one antenatal care (ANC) clinic at 36 weeks. Following an unsuccessful mixed management of the third stage of labor (MMTSL) and a failed manual removal of the placenta, she underwent a laparotomy and hysterotomy indicated for failure of manual removal of the placenta with PPH. Intraoperatively, the placenta was firmly adhered to a large non-pedunculated submucosal uterine myoma. The myomectomy done led to the extraction of a myoma with a complete placenta adhered to it. The myoma measured 15 x 5.8cm and the fetus weighed 2800g. The histopathological examination of the placenta and myoma was not done due to financial constraints and lack of technical platform. The diagnosis of placenta accreta was therefore not confirmed but the myoma and placenta were successfully extracted; and the uterus was preserved. This potentially preserved the patient's chances of future conception.

Key Clinical Message

Cases of retained placenta due to placental implantation to a submucosal uterine myoma are quite rare, yet the danger they represent is tremendous. First and second trimester ultrasound are key in the early identification of such cases, however, few pregnant women only in low-resource settings do it, which calls for concern.

Keywords

Case report, Leiomyoma, Placenta Accreta, Placenta, Retained, Pregnancy.

Introduction

Leiomyomata occur in 0.1–10.7% of all pregnancies worldwide, being more common in primigravidae due to their previous state of nulliparity [1]. Complications associated with leiomyomata in pregnancy are not uncommon, with placental abnormalities being more and more reported [2]. It is not uncommon for pregnant women to have a placental implantation on an underlying uterine myoma. However, for this phenomenon to happen, there should be a clear and established connection between the placenta and the myoma through mechanisms like direct contact from pedunculation and degenerative changes of the myoma, promoting the formation of adhesions between the placenta and the underlying myoma [3]. Therefore, an adherent placenta should be expected

in cases of submucosal leiomyomata especially the pedunculated forms [3]. We are reporting the case of a retained placenta adhered and implanted to a large non-pedunculated submucosal myoma observed at the Ekona Subdivisional Hospital in the Southwest region of Cameroon.

Case History

Patient information

She is a 31-year-old pregnant woman, of a local tribe, G1P0, blood group A rhesus positive, who presented at 39 3/7 weeks' gestation with intermittent and crampy lower abdominal pain of gradual onset that started 24 hours prior to presentation. There was no fever, no gush of fluid and no vaginal bleeding. The pain increased in frequency and intensity with time, from experiencing a pain of 4/10 on the verbal numerical scale (VMS) every 2 to 3 hours to a pain of 7/10 every 1 to 2 hours. The latter eventually prompted her consultation at our services (Ekona Subdivisional Hospital).

For this pregnancy, her booking visit and only antenatal care (ANC) visit was at 36 weeks of gestation. At the booking visit, her haemoglobin level was 10.8 g/dL and she started taking 0.04 mg of folic acid daily and 60mg of elemental iron daily. She received 2 doses of sulphadoxine-pyrimethamine (SP) for malaria prevention. A single ultrasound scan was done at the booking ANC visit and reported a normally evolving single viable intrauterine pregnancy at 36 weeks of gestation, with no evidence of a myoma. She has no history of hypertension or diabetes mellitus before the current pregnancy. She denies any family history of the aforementioned diseases.

Clinical Findings

On physical examination, she was conscious and alert, screaming in pains and tachypneic. Her blood pressure was 118/70 mmHg, pulse rate of 106 beats per minute, respiratory rate, was 24 breaths per minute, oxygen saturation (SaO₂) was 100%, and axillary temperature, was 37.2°C. Her conjunctivae were pink and the sclerae anicteric. On cardiac auscultation, the first and second heart sounds were heard and normal on all four cardiac auscultatory areas. Her lungs were clear. Her abdomen was distended with a gravid uterus in line with her gestational age. The symphysio-fundal height (SFH) was 30cm and abdominal circumference (AC) was 95cm. The lie was longitudinal and presentation was cephalic. The foetal heart rate (FHR) recorded was 138bpm. The cervix was posterior, medium, 50% effaced and 4cm dilated. Station was -1.

Diagnosis: Latent phase of the first stage of labour in a primid.

Timeline of the current episode

She was admitted in the maternity unit and a partogram was opened. Spontaneous rupture of membranes with gush of clear amniotic fluid occurred 30 minutes after admission. The labour progressed to the spontaneous vaginal delivery of a life male baby whose APGAR score was 8 at the 1st minute and 10 at the 5th minute. The baby weighed 2800g.

Following the delivery of the baby, attempts to extract the placenta through the active management of the third stage of labour were unfruitful. During the latter, there was complete rupture and detachment of the cord from the placenta during control cord traction and eventually postpartum hemorrhage (PPH). The cord was thin and friable. This was followed by several unsuccessful attempts to expel the placenta and control the bleeding using uterotonics. These attempts included a total of 50IU of oxytocin administered intravenously (IV) (40IU in 500cc of Normal saline administered at a flush rate and 10IU intramuscularly) and 200 micrograms each of misoprostol sublingually and intrarectally (IR). After failure of medical expulsion, manual removal of the placenta was done. During the manual removal attempts, it was noticed that the placenta was firmly attached to the endometrium without any possibility of separation at the line of cleavage. Attempts to expel/extract the placenta both medically and manually after an hour and 15 minutes were unfruitful. Thus, a working diagnosis of retained placenta with PPH, probable etiology being placenta accreta was made.

Therapeutic interventions

She was then prepped and transferred to the theatre for a laparotomy and hysterotomy indicated for failure of manual removal of the placenta with PPH. This was done in an attempt to surgically extract the placenta and control the PPH while keeping a high index of suspicion for placenta accreta, since other PPH-causing factors like uterine atony and trauma were absent. Intraoperatively, a fully-formed placenta was visualized firmly adhered to the posterior uterine wall with a difficultly identifiable line of cleavage. The endometrial lining was irregular around the placenta, appearing elevated together with the placenta. Further examination unveiled the presence of a firm round mass underneath and around the placenta. An incision was done along the line of cleavage and a non-pedunculated submucosal myoma was visualized firmly adherent to the placenta. The myoma was delicately extracted together with the placenta with no further attachment to the myometrium. The myoma measured 15 x 5.8cm, and the placenta was complete with all cotyledons present (Figure 1). Hemostasis was done and checked several times before final closure of the uterus and abdominal wall. Estimated blood loss was estimated at 800cc, and urine output was 600cc. The surgery was done under general anaesthesia. The lack of technical platform did not permit histopathological examination of the placenta and myoma.

After the surgery, she received intravenous (IV) fluids 2500 ml/24 hour on postoperative day 0 through day 1. In addition to that she received Ceftriaxone 1 g/12 hourly (IV) on postoperative days 0, 1 and 2. She also received tramadol 100mg (slow IV) 12 hourly alternating with diclofenac 75mg (IM) 12hourly from postoperative day 0 to day 2. Oral relay was done on postoperative day 3 with tramadol-paracetamol (325mg+37.5mg) combination 1 tablet TID for 5 days, amoxicillin-clavulanic acid (875mg+125mg) combination 1 tablet TID for 7 days and methylethergometrine 0.125mg, 1 tablet TID for 3 days.



Figure 1: Picture of extracted placenta (up) and myoma (down) with contour (outline) of the myoma imprinted on the maternal surface of the placenta.

Follow-up and outcome of interventions

Following the surgery, the recovery was uneventful after 8 days of admission. By postoperative day seven, she had sustained major clinical improvement, her baby was clinically well and both were discharged. Discharge criteria for the mother included; minimal postoperative pain, absence of fever, lochia serosa and steady uterine involution rate (SFH of 30cm on entry and 23cm on discharge).

Discussion

It is worth to note that pregnancy in several ways has a deep influence on leiomyomata. Some of these influences include red degeneration, necrosis, infection, torsion of pedunculated leiomyomata and in several cases an increase in leiomyomata' volume in 20–30% of cases [4].

Leiomyomata in pregnancy are usually diagnosed in 4–5% of women undergoing prenatal ultrasound and first trimester ultrasound [4]. Evaluation of pregnancies in the first trimester, makes the diagnosis of associated leiomyomata easier, with the uterus in the pregnancy being small, thus easier to examine fully. The detection and evaluation of leiomyomata as the pregnancy progresses is difficult due to changes in uterine anatomy and the presence of the fetus and placenta that sometimes make examination

difficult. However, the detection remains quite feasible in the early and mid-second trimester, thus making this period also suitable for ultrasonographic diagnosis of leiomyomata in pregnancy and its relation to the placenta [5]. It therefore becomes challenging to establish the diagnosis of a uterine myoma in the later trimesters of pregnancy (late-second and third trimesters), becoming more and more challenging as the pregnancy progresses. This becomes even more troublesome in low- and middle-income countries (LMICs) where most pregnant women are known to start antenatal care (ANC) visits and do ultrasounds late (usually in the third trimester) like for the index case [6]. Prenatal, first and second trimester ultrasounds are therefore the gold standard to diagnose leiomyomata in pregnancy and possible adhered placentae to leiomyomata.

Women with leiomyomata are more likely to experience third-stage labor difficulties and puerperal problems, and they are more likely to have a retained placenta [7]. This is because, they tend to have dysfunctional labors as a consequence of the interference of myometrial contractility, and as a result their placentae are less likely to detach from the endometrium increasing the chances of a retained placenta and therefore postpartum hemorrhage (PPH). They therefore are more likely to use uterotonics to control uterine contractions and to increase the chances of successful placenta separation (detachment) [7].

Submucosal leiomyomata are the least common type of leiomyomata (5%) with the pedunculated submucosal type accounting for only 2.5% [3]. However, despite being the least common, pedunculated submucosal leiomyomata can interfere with implantation causing infertility or they can cause miscarriage or preterm labour. They can also interfere with placentation giving rise to various forms of abnormal placentation or placental abnormalities [8] such as in recent data showing a 3-fold increase in the risk of abruption placentae and a 2-fold increase in the risk of placenta praevia in pregnant women with leiomyomata, especially if these leiomyomata have a volume of more than 200 cm³ and if they are retroplacental or submucous in location [3,8]. Nevertheless, in certain cases, the placenta can adhere to the uterine myoma resulting in an adherent placenta because of its implantation on the myoma. This could result in placenta accreta, which is an obstetrical catastrophe [3]. In that light, submucosal leiomyomata have long been recognized as one of the causes for placenta accreta [3]. Again, for this event to happen, there should be a connection between the placenta and the myoma through mechanisms like direct contact from pedunculation and degenerative changes of the myoma, promoting the formation of adhesions between the placenta and the underlying myoma. An adherent placenta would therefore be expected in cases of submucosal leiomyomata especially the pedunculated forms [3]. This was the case with Dasari's report of a case of placenta accreta associated with submucosal fibroid polyp in India where a pedunculated fibroid polyp was found adherent to the placenta of a pregnant woman at 33' 4/7 weeks of gestation. The fibroid polyp was attached to the placenta by a peduncle that connected both structures. It is therefore rare to observe cases of

adhered placenta to non-pedunculated leiomyomata, especially in primigravidae, without any invasive uterine procedure done in the past that could create a connection between the endometrium and the deeper layers as it was the case for the index case (Figure 1). It is also worth to note that, in majority of cases of placenta accreta, hysterectomies remain the most popular option used by health professionals due to the inability to extract the placenta indefinitely and thus, the terrible complications that may ensue from that. However, despite the life-saving benefits of such a procedure for cases of placenta accreta, the implications in primigravidae especially the advanced ones remain tremendous, with their chances of conception being taken away by the surgery [9]. This would equally apply to cases of adhered placenta to leiomyomata due to the failure of placental removal and the risk of development of intractable postpartum hemorrhage and placenta accreta. Therefore, avoiding hysterectomy when possible in these cases could salvage their chances of conception and thus improve their quality of life.

Surgical treatment of leiomyomata is uncommon in pregnancy, particularly during the first trimester [10,11]. The use of procedures such as using 200 mg of misoprostol sublingually and intrarectally, and/or oxytocin infusion or intravenous administration of Carbetocin in bolus during CS have been proven to minimize blood loss [10]. Although conservative management is considered the gold standard, in some cases surgery must be performed. However, if the placenta becomes adherent to a large myoma and ends up being retained, conservative management as well as manual removal of the placenta are likely to be unsuccessful [10,11]. Therefore, surgical management would likely become the sole option, presenting either as a myomectomy during a lower uterine segment caesarean section (LSCS), as a laparotomy following a vaginal delivery as for the index case (Figure 1) or as a hysterectomy. The latter remains the least favorite option as it will involve the removal of the uterus together with their chances of future conception.

Placenta accreta is a very dangerous obstetric complication associated not only with high maternal mortality rates but also with drastic decrease in fertility rates due to the fact that its management is mainly through hysterectomy, especially in LMICs [12]. The rate-determining step in this case therefore is the diagnosis of placenta accreta due to its paramount implications on the fertility of the mother. The diagnosis of placenta accreta is primarily histopathological, which poses a great challenge to achieve in LMICs due to lack of suitable technical platform for that purpose [13]. In such settings, the management of cases of retained placenta refractory to medical and manual management especially in the setting of PPH, very often lead to a presumptive diagnosis of placenta accreta and a subsequent hysterectomy being done [14].

However, in rare cases like for the index case, the retained placenta could rather be a placenta adhered to a myoma like for the index case (Figure 1). In such cases, the extraction of the placenta will

follow the myomectomy, thereby avoiding a hysterectomy and thus, preserving the mother's chances of conception [3].

Myomectomy is of proven benefit and in such cases even more because it does not impair maternal chances of conception as observed in a study carried out in 2006 in Italy, where patients who underwent myomectomy for resection of submucosal fibroids had higher clinical pregnancy rates when compared to patients with fibroids that did not undergo surgery. The likelihood of live births and spontaneous abortions were similar in both groups, also suggesting that clinical pregnancy, live birth, and spontaneous abortion rates will normalize over time in women with submucosal fibroids following myomectomy when compared to infertile women without fibroids [15]. However, even though it does not impair the chances of conception, it rather increases the chances of placenta accreta spectrum disorders as observed in a United Kingdom-based nationwide cohort study carried out in 2012, that found out that myomectomy is associated with an increased risk of PAS in the subsequent pregnancy. In addition, among different methods of myomectomy, hysteroscopic myomectomy was correlated with a significant higher risk of PAS in the following pregnancies than laparotomic or laparoscopic myomectomy. Surgeons should consider this and inform all patients who are to undergo a myomectomy about the potential implications on future pregnancies [16].

Conclusion

Leiomyomata in pregnancy especially the submucosal forms are associated with several complications, notably abnormalities in placentation. One of the most dangerous yet common placental abnormalities associated with this form is placenta accreta due to the adherence and implantation of the placenta to the submucosal myoma. This phenomenon poses a great challenge both in the diagnosis, requiring prenatal, first or second trimester ultrasound(s) to be diagnosed, and in the management, requiring surgical removal which in pregnancy is not usually envisaged because of the bleeding risk it poses. Furthermore, the clinical similarities between adhered placenta and placenta accreta; and the need for histopathological confirmation of placenta accreta make the decision-making regarding the management of such cases very challenging, taking into consideration the high rates of hysterectomy associated with placenta accreta which in primigravid women like the index case could significantly and negatively impact their future fertility. The management of cases of adherent placenta to a myoma like for the index case remains extremely challenging based on all what is at stake especially for the mother's survival and future fertility. Moreover, it becomes even more challenging in LMICs where few pregnant women do first or second trimester ultrasounds, few attend ANC clinics, and where in primary health facilities like for the index case, there is a limited technical platform to explore all the management options. Early diagnosis, interdisciplinary collaboration and improvement of tools to predict and diagnose both abnormally invasive and abnormally adherent placenta are key in preventing this rare condition.

This case illustrates the outcome of a pregnancy when the placenta was retained following a vaginal delivery because it was implanted (adhered) on non-pedunculated submucosal uterine myoma.

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