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Case Report

Successful outcome of a triplet pregnancy following laparoscopic myomectomy for infertility: a unique case report

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ABSTRACT

Infertility is a burning problem affecting 10-15% of the couples globally. The female factors contribute to about 40-45% among which uterine factor is up to 10%. Presence of uterine myomas also significantly contribute to infertility. A conservative surgical approach like myomectomy is indicated for women in reproductive age both for psychological reasons as well as to preserve their reproductive potential. In recent years laparoscopic myomectomy (LM) has become the procedure of choice. Pregnancy outcome after myomectomy is a more significant concern especially with regard to the obstetric calamity of sudden uterine rupture due to the presence of an operative scar. The intensity of such complication will be considerably high when a triplet pregnancy occurs with a LM scar in the upper uterine segment with history of uterine cavity being opened during the myomectomy surgery. Managing such a case will be a difficult task to the attending clinician posing many unexpected clinical dilemmas. We are reporting a unique case of triplet pregnancy occurred following a LM resulting in successful maternal and fetal outcome. During the course of this pregnancy apart from considerable maternal distress due to uterine over distension many clinical problems like cervical insufficiency, pre eclampsia, polyhydraminos, intrauterine growth restriction (IUGR) and last but not the least severe postpartum haemorrhage (PPH) have been encountered and could be managed efficiently. An elective Caesarean section was performed at 33^{+4} weeks gestation delivering live triplets. Mother and all the three babies discharged from the hospital in good condition.

Keywords: Infertility, Laparoscopic myomectomy, Triplet pregnancy

INTRODUCTION

Couples presenting with an inability to conceive need special attention of a gynaecologist and sometimes a psychiatrist since the problem is complex. As infertility causes lot of distress and emotional problems leading to even family disruption at times, such couples have to be handled with sensitivity and understanding. Globally the prevalence of infertility is about 10-15% in couples of reproductive age and it increases with increasing age of the females in particular. The female factors for infertility contribute up to 40 -55% out of which uterine factor is to an extent of 10%. Among the uterine factors uterine leiomyomas also significantly contribute to female infertility.[1]

Conservative surgery in the form of myomectomy for women in reproductive age is ideal so as to preserve their fertility potential.[2] In recent years LM has become the elective procedure in selected patients (a single intramural/sub serous myoma).

Isolated case reports of uterine dehiscence/rupture have been published in the medical literature, suggesting that the risk of this complication may not be negligible.[3,4] The anticipated pregnancy complications will be of higher

frequency and intensity in multi fetal pregnancies especially of the higher order (Triplets onwards) occurring with Lm scar in the upper uterine segment, (particularly with H/o of cavity being opened during surgery and deep penetrating myomectomy wound). Therefore, the outcome of such a pregnancy is of significance.[3]

CASE REPORT

A 29 years aged woman teacher by occupation married and sexually active for three years attended the outpatient department of Narayana medical college and hospital (NMCH) a tertiary care centre, Nellore, Andhra Pradesh, India, with the complaint of not being able to conceive despite having had medical treatment in various hospitals for the last three years. As per the history her in laws were beginning to worry and the pressure was taking its toll on their relationship. All in all, it was a bit too much to bear and the couple were confused and anxious.

Her menstrual history and coital history are normal. Husband teacher by occupation, not a smoker, no addiction to alcohol/drugs. Semen analysis: WNL. No other relevant history.

On clinical examination: Moderately nourished. BMI: 19kg/m^2; vital signs: WNL; Thyroid and Breast: normal; P/A: A mid line swelling felt corresponding to 14 -16 weeks of pregnancy, mobile and firm; P/S: no abnormal findings; P/V - Uterus enlarged to 14-16 weeks, feels irregular.

Investigations: Thyroid profile, FSH (day 3), Serum prolactin: WNL; HSG (done outside in 2013) - no intra uterine filling defects seen. Peritoneal spillage observed from both tubes; Ultra sonogram (USG) - Enlarged uterus with well defined heterogeneously hypo echoic lesion measuring 6.5 x 8 cms in anterior myometrium with mild distortion of the endometrial cavity; Diagnostic Hysteroscopy - the Anterior fibroid appears to be projecting into the uterine cavity to an extent of 20 – 25%. Visualization of tubal Ostia was slightly difficult due to the myoma's projection in to the upper part of uterine cavity. The final diagnosis of anterior intramural myoma (single) was made and couple counselled about the treatment options. The couple with their acquired knowledge through internet opted for LM. Case posted for laparoscopic myomectomy under G.A after the required work up.

Intra operative findings

- Single anterior myometrial fibroid of about 8 x 7 cms, the prominent part being noticed just below the fundus and tubal cornua.
- Right ovary normal and left ovary cystic.
- Before proceeding with the surgery chromotubation was done to make sure of the tubal patency. Both tubes: patent.

Following vasopressin injection an incision is given in the most prominent part of the myoma with the monopolar spatula up to the pseudo capsule. Myomectomy was performed with aqua dissection and sparingly using the bipolar for haemostasis. During the enucleation of myoma the uterine cavity was found to be opened and the same repaired with 0 - vicryl. Myometrial closure was done with 1- vicryl taking the full depth of the incisional edges so as to avoid secondary constitution of a haematoma deep inside the myometrium. Haemostasis verified and about one litre of ringer's lactate solution was left in the peritoneal cavity to potentially reduce adhesion formation. Post-operative period was uneventful.

She returned within 6 months asking for medication to conceive due to the pressure of her in laws. She was asked to wait for few more months. After a month she resurfaced again at the hospital with history of missed period and a positive pregnancy report. Eight weeks later the dating scan revealed a triplet pregnancy to the utmost surprise of everyone. The couple again did their part of research through the internet and understood the risk of miscarriages etc., in such multiple pregnancies. They are also found to be aware of selective termination and the associated risks. Finally, they have decided to continue the pregnancy as it is with the firm determination to face what may come. Understanding their problems, she was registered as a high risk pregnancy and ante - natal booking was done.

Routine ante natal investigations were done: WNL; She was advised Tab. Folic acid and twice weekly inj. Hucog (5000 I.U.) till 16 weeks.

USG at 12 weeks: Nuchal translucency: WNL; Trichoronic triamniotic triplet pregnancy; TIFFA scan (22 wks): normal study.

At 23 weeks she came to the hospital with the c/o of back pain. USG: revealed cervical shortening (2cms) along with funnelling observed. Clinical examination confirmed the scan findings - Cervical encerclage done after counselling the couple.

At 28 weeks pregnancy: she started complaining of slight breathlessness. O/E: Pedal edema +; BP: 130/90 mmHg; P/A: Fundal height was corresponding to 32 wks; urinalysis: proteinuria 1+. She was advised admission for observation and investigations (sr. Creatinine: 0.8mgn %, Coagulation profile and LFT: WNL .24 hrs urine protein value - 0.3gms.

At 30 weeks she still feels breathless and Bp readings were ranging between 130-140 / 90-100 mmHg. She was started on Tab. Labetalol 100 mg BID.USG: revealed normal well being of all the three foetuses.

At 32 weeks she is still in the hospital under observation; Bp: 140/90mmHg (with tab Labetalol 100mg BID);

Fundal height is almost 36 weeks with tense abdominal wall; USG: Polyhydraminos +, there is a growth lag of 1-2 wks observed in all the three babies; Doppler velocimetry: Umbilical artery S/D increased, end diastolic flow absent; MCA: WNL; Biophysical prolfile:8/10

At 33 weeks 2 days with further increase in breathlessness and uterine over distension being increased decided to terminate the pregnancy by elective Caesarean section (fear of impending scar rupture). Doppler velocimetry same findings as before. Advised and given two doses of inj. Betnesol.

At 33 weeks 4 days elective Caesarean section was performed (4 units of compatible blood being reserved in anticipation of PPH). Cervical stitch removed in the OT.

Intra operative findings

- Lower uterine segment was found to be thin and well formed.
- All the three babies are delivered with apgar scores of 6-8 and birth weights ranging from 1.6 to 1.9kgs.
- Placentas delivered in toto (Trichoronic triamniotic).
- Scar in the upper segment of the uterus was felt to be papery thin, but intact.
- As anticipated there was severe atonic post partum haemorrhage which could not be well controlled with step wise uterine devascularisation. Finally, uterine packing was done.
- General condition was fair, and two units of packed cells were transfused in the theatre.

Post operative period uneventful. Uterine pack was removed slowly after 36 hours. Mother and babies discharged in healthy condition.

Figure 1: Ultrasound image: triplet pregnancy.

DISCUSSION

Infertility is a burning problem affecting 10-15% of the couples. 40 - 50% contribution is by the female partner and 10% by the uterine factors. Uterine myomas are the commonest pelvic tumours found in at least 20% of the females over thirty years of age. Their presence also significantly contributes to infertility and pregnancy failures. A conservative surgical management include myomectomy for women who wish to retain their uterus for continuing their reproductive potential.[5] Laparoscopic myomectomy with all its advantages can be offered to these patients.

Laparoscopic myomectomy- reproductive performance

The patient satisfaction with operative scar after LM is good along with fertility and reproductive outcomes. The literature documents normal reproductive performance of uteri after LM (Li et al). Conception rates have been quoted to be 71% and 75% vs 40 and 58 % in abdominal myomectomy in two different series.[6] Provided no other associating factor for infertility is found, LM enhances fertility rate.[7] Paul et al showed that the majority of their patients conceived in the first year after surgery (82.6%) and a significant number in the first six months (55.6%).[8] Present case also conceived in the seventh post operative month indicating that removal of the fibroid enhanced her fertility potential as there is no other plausible infertility factor.

Triplet pregnancy- reproductive performance

Multifetal pregnancies are high risk pregnancies. Pregnancy complications along with maternal and fetal risk increase hand in hand with higher order multiples. The various pregnancy complications being Anaemia, Preeclampsia, IUGR, Preterm deliveries, PPH and even post natal depression. The most common complication is preterm birth more than a half of all twins are born preterm and higher order multiples are almost always preterm. In a review of 19 sets of triplets the most common antenatal complications were preterm labour and preeclampsia. The mean gestation at delivery was 33 weeks (range 25-39 weeks).

Multi-foetal pregnancies (triplets onwards) of the higher order constitute high risk groups and contribute to the higher rate of childhood mortality especially during early period of life. Children born in these cases were more than twice as likely to die during infancy as infants born singleton.[9] In our case apart from the complications of preeclampsia, prematurity, and IUGR additionally Cervical insufficiency, polyhydramnios with considerable maternal pressure effects (breathlessness) have been encountered.

Atonic PPH in our case can be attributed to the uterine over distension caused by triplets perse along with polyhydramnios resulting in uterine atony. It is well anticipated and taken care of. Post natal depression was also expected but on the contrary the couple were extremely happy to have three babies at a time after a long period of awaiting.

Laparoscopic myomectomy - uterine scar dehiscence /rupture:

Several studies have evaluated the pregnancy outcome among patients who underwent myomectomy which is a challenging procedure because it involves the reconstruction of an organ that can undergo remarkable structural changes, as it does in pregnancy. In recent years LM has become the elective procedure in selected patients. Laparoscopy effectively shortens the hospital stay and avoids the major risk of the classical route i.e. adhesion formation (Bulletti et al, Dubuisson et al.). Questions have been raised about how this new approach affects the outcome of pregnancies following surgery (Dubuisson et al, Darai et al, Nezhat et al, Ribeiro et al) making pregnancy outcome after LM as a more significant concern.[10] Because myomectomy is often performed to preserve the uterus for future pregnancy, maintaining the integrity of the uterine wall is of utmost importance (Dubuisson et al).

Isolated case reports of uterine dehiscence in pregnancy have been published in the medical literature, suggesting that the risk of this complication may not be negligible.[2] The prevalence of uterine rupture following myomectomy (0.79%) is comparable with that after Caesarean section. Based on the available evidence, there is no statically significant difference between the incidence of scar rupture during pregnancy following a laparoscopic versus an open myomectomy.[11] In selected cases, LM has been reported to be an effective technique that is associated with a low rate of patient morbidity (Dubuisson et al, 1996).

One possible cause of uterine rupture after LM is the wide use of thermal energy which may result in poor vascularisation and tissue necrosis with an adverse effect on scar strength (Elkins et al, Nezhat et al). Thermal energy was used to remove the myoma and obtain haemostasis in five out of the six reported uterine ruptures. As far as suturing is concerned, the problem is the correct reapproximation of the edges of incision so as to prevent the formation of haematoma deep in the myometrium. This precaution is necessary to diminish the likelihood of healing by secondary repair.[10] When the uterine cavity has been opened or when the myomectomy defect is deep, it is necessary to make a suture in two planes. In our case as the uterine cavity was found to be opened during the procedure of LM it was separately sutured followed by suturing and good approximation of the myometrial edges taking all the above mentioned precautions. Our judicious use of thermal energy during the procedure also must have contributed to the good scar integrity thus preventing scar dehiscence / rupture till the time of delivery.

Triplet pregnancy - uterine scar dehiscence /rupture

Compared to singleton pregnancy the incidence of scar rupture will be high in multiple pregnancies especially of the higher order multiples due to associating over distension and other pregnancy complications. Since our case being a triplet pregnancy and LM scar being in the upper part of the uterine body with history of cavity being opened the chances for such calamity was much feared but it did not happen probably due to the precautions that were taken while suturing myomectomy wound.

Laparoscopic myomectomy - risk of operative delivery

Previous myomectomy and/or infertility were indications for almost half of the elective Caesarean sections performed. Though these are not the absolute indications for the abdominal route worries by both patients and their Obstetricians could play a more decisive role than actual pathological indications.[10] If this is the case in singleton pregnancies how triplets should be delivered with previous LM scar? Some studies indicate a significantly improved perinatal mortality for the second and third babies when delivered by Caesarean section. Factors that increase the perinatal loss during vaginal delivery are low birth weight and prematurity, delay between delivery of each fetus, poor monitoring of second and/or third fetus during labour, and manipulative procedures during delivery thus the later-born infants are especially at risk. In contrast to this view, Itzkowic considered vaginal delivery as reasonable after 34 weeks' gestation in uncomplicated triplet pregnancies. However, in his review of 59 triplets all 27 babies delivered by Caesarean section survived, while the 9 stillbirths and 32 neonatal deaths all followed vaginal delivery.[12] In another review of 19 sets of triplets the outcome of triplet pregnancies was better in the group that delivered vaginally than those delivered by Caesarean section. Greater maturity of the infants delivered vaginally appeared to be the major factor for the lower neonatal morbidity and mortality. Nezhat et al described that the increased incidence of Caesareans is not surprising, since this is the recommended method of delivery for women in whom the uterine wall has been deeply penetrated during myomectomy.[13]

Current case presents a unique scenario with a long period of infertility, LM scar in upper segment (akin to hysterotomy scar) with the cavity being opened during that procedure, multifetal pregnancy of the higher order (Triplets) and along with the various pregnancy complications that have been encountered made elective Caesarean section mandatory in this case.

CONCLUSION

LM is no doubt an ideal procedure for infertile women of reproductive age. This is especially feasible when there is a single intra mural myoma. The surgeon's experience in laparoscopic suturing is a crucial requirement for smooth outcome of both LM and subsequent pregnancies. Appropriate patient selection is particularly important for infertile patients, in whom postoperative adhesions and

uterine integrity are key factors in conceiving a child and carrying it to term.

All the enumerated pregnancy complications with LM scar may be encountered even in singleton pregnancies. Our case is different in various aspects. Apart from the inherent risks that associate with triplet pregnancy the other important risk factor in this case is the LM scar from previous surgery. Combination of operative scar in the upper uterine segment with accompanying over distension of the uterus by the triplet pregnancy are the worrying factors about the successful pregnancy outcome. All the problems encountered during the journey from conception to delivery could be effectively managed and a timely decision for Caesarean section resulted in a fruitful outcome of bringing live triplets in to this world.

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