Utero-Cutaneous Fistula Following Caesarean Section Secondary to Degeneration of Fibroid: A Rare Case

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Abstract: Uterocutaneous fistulae are extremely rare compared to common cutaneous fistulae involving ureteric and gastrointestinal tracts. Uterocutaneous fistula is a rare complication following caesarean section with only a few cases published in literature. This is the second case report of utero-cutaneous fistula where the communication is between the endometrial cavity and skin lesion via a necrotic intramural fibroid following caesarean section (CS). A primigravida underwent classical caesarean CS in view of large lower segment intramural fibroid. She also underwent step wise devasularization for intractable PPH. After two months of initial CS she reported back with pus like discharge at the incision site. The diagnosis of utero-cutaneous fistula was made which was confirmed by MRI. At laparotomy, fibroid necrosis and fistula tract was seen. Total abdominal hysterectomy with excision of fistulous tract was done. Histopathologic examination revealed intramural leiomyoma with extensive infarction. The case highlights this rare complication in uterine fibroid and caesarean section. Once a fistula is diagnosed, prompt timely intervention with excision of the fistulous tract is warranted.

Keywords: Utero-cutaneous fistula, fibroid, fibroid necrosis, obstetric fistula, fistula

1. Introduction

Fistula is an abnormal communication between two epithelial surfaces. Vesicovaginal and rectovaginal fistulae are common genital fistulas well known in gynaecology whereas uterocutaneous fistulae are very rare. Fistulae involving the uterus are usually result of abortions, postpartum and postoperative complications. This is the second case report of uterocutaneous fistula where the communication is between the endometrial cavity and skin lesion through a necrotic intramural fibroid following caesarean section.

2. Methodology/ Case Report

A 28 year old primigravida was referred as fibroid uterus complicating pregnancy to our tertiary care hospital. Dating scan at 7 weeks showed multiple fibroid uterus with largest in anterior wall lower uterine segment measuring 17×15×12cm and another large lateral wall fibroid measuring 10×8×6cm. When she presented to us at 12 weeks of gestation, her fundal height was 20 weeks. This was further complicated by major degree placenta previa and the fibroid had further grown in size to 25×21×16cm by 32 weeks.

She presented with bleeding pervaginum at 33 weeks and underwent emergency classical CS in view of large obstructing lower segment intramural fibroid with placenta previa with antepartum haemorrhage. A live baby of 1.7kg with good APGAR was extracted. During the surgery she developed intractable PPH which was controlled by step wise devasularization with bilateral uterine artery, utero ovarian artery ligations including bilateral internal ileac artery ligation. Her postoperative period was unremarkable and was discharged on post op day 8.

After two months of initial CS she reported back with pain abdomen and pus like discharge at the incision site through a small opening. She also complained of similar discharge from vagina. There was no history of fever. The diagnosis of utero-cutaneous fistula was made which was confirmed by MRI. At laparotomy, fibroid necrosis and fistula tract was seen. Total abdominal hysterectomy with excision of fistulous tract was done. Histopathologic examination revealed intramural leiomyoma with extensive infarction. The case highlights this rare complication in uterine fibroid and caesarean section.

Figure 1: MRI axial view
3. Discussion

Utero-cutaneous fistula is a rare clinical entity with very few cases reported worldwide in literature till date. Uterocutaneous fistula usually results from post abortion, post-partum or postoperative complications. Other causes include endometriosis, tuberculosis and Crohn's disease while others may bedue to tumour infiltration. Gupta et al reported first case of uterocutaneous fistulain 1993 which developed following septic abortion induced by laminaria tent insertion in the cervix. Dragoumis et al in 2004 described a case of endometriotic cutaneous fistula after CS. Promosonthi et al in 2007 described another case of uterocutaneous fistula secondary to an abscess caused by in situ left placenta after an abdominal pregnancy. Eldemet al in 2008 reported a case of uterocutaneous fistula developed secondary to CS performed 19 years ago. Baggish et al in 2010 reported another case of uterocutaneous fistula as a complication of ruptured appendix and Crohn's disease during pregnancy. Pant et al reported in 2012 a case of uterocutaneous fistula of tubercular etiology following CS. Lim et al reported a case in 2012 , utero-cutaneous fistula due to red degeneration of intramural fibroid where they did classical CS for retained second twin.

Uterocutaneous fistula is a rare condition that may be difficult to manage. Surgical excision of the fistulous tract is the treatment of choice. Most cases end up with hysterectomy however Seyhan et al reported a case of uterocutaneous fistula that was successfully treated with gonadotropin-releasing hormone agonist administration. We hypothesise that, in our case the cause for fistula could be impaired vascular supply to large fibroids after internal iliac ligation. Burbank and colleagues put forward this ‘transient uterine ischemia’ hypothesis, to explain the mechanism of uterine artery occlusion. They proposed that after uterine artery occlusion, both the myometrial and myoma vessels were occluded by clotting, resulting in organ ischemia. The metabolism shifts from oxidative pathways to anaerobic glycolysis. The study on uterine artery occlusion found that necrosis in the myoma, but not in the myometrium, contributed to the shrinkage of tumors, which was shown both through pathological observations and with MRI.

4. Conclusion

This is the second case report of utero-cutaneous fistula where the communication is between the endometrial cavity and skin lesion via a necrotic intramural fibroid following caesarean section. This case highlights the rare complication in uterine fibroid and caesarean section. This also emphasises the risk of increased morbidity when we opt for conservative approach in the presence of very huge multiple fibroids. Once a fistula is diagnosed, prompt timely intervention with excision of the fistulous tract is warranted.
References


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