

# 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 devascularization 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.<sup>1,2</sup> 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 pervaginam 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 devascularization with bilateral uterine artery, utero ovarian artery ligations including bilateral internal iliac

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 which showed fistulous tract between uterine fibroid and skin with large collection around. (fig1,2) She was readmitted and underwent emergency laparotomy. Intraoperatively it revealed extensive necrosis of fibroid with fistulous communication between skin and endometrial cavity through necrotic intramural fibroid. The necrotic material presented as a collection around the fistulous tract and was draining out both through the fistula percutaneously and from endometrial cavity through vagina. (fig3,4) Total abdominal hysterectomy with excision of fistulous tract was done. She was treated with broad spectrum antibiotics and recovered with uneventful postoperative period. Histopathological examination revealed intramural leiomyoma with extensive infarction. (fig5)

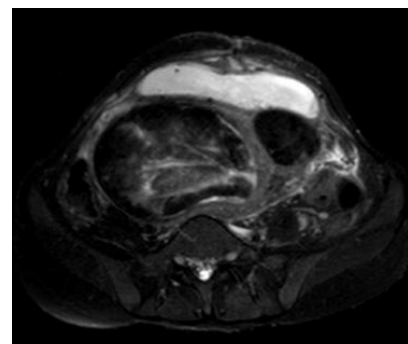
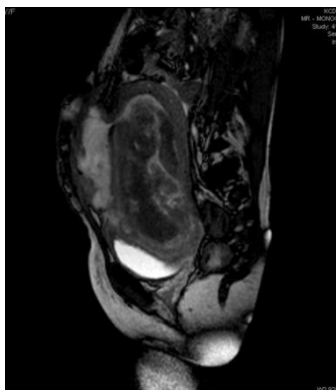
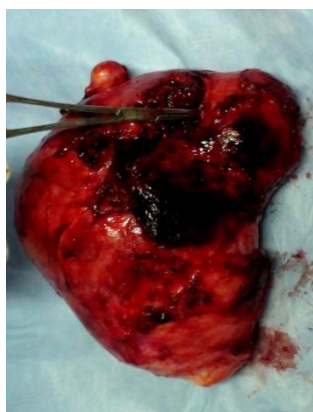


Figure 1: MRI axial view



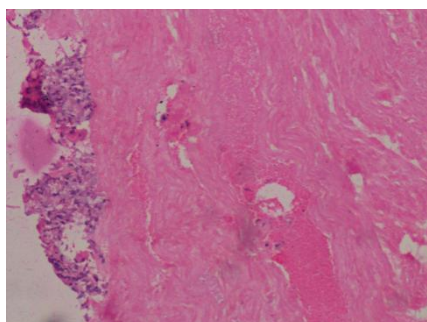
**Figure 2:** MRI Saggital view ( fig 1&2 show degenerated with cutaneous fistula)



**Figure 3:** Hysterectomy specimen fistula being pointed



**Figure 4:** cut section of uterus showing necrosed fibroid



**Figure 5:** Histopathology – intramural leiomyoma with extensive infarction

### 3. Discussion

Utero-cutaneous fistula is a rare clinical entity with very few cases reported worldwide in literature till date.<sup>1</sup> Uterocutaneous fistula usually results from post abortal, post-partum or postoperative complications. Other causes

include endometriosis, tuberculosis and Crohn's disease while others may be due to tumour infiltration.<sup>2-9</sup>

Gupta et al<sup>2</sup> reported first case of uterocutaneous fistula in 1993 which developed following septic abortion induced by laminaria tent insertion in the cervix.

Dragomirescu et al<sup>3</sup> in 2004 described a case of endometriotic uterocutaneous fistula after CS.

Promosonthi et al<sup>4</sup> in 2007 described another case of uterocutaneous fistula secondary to an abscess caused by in situ left placenta after an abdominal pregnancy.

Eldemet al<sup>5</sup> in 2008 reported a case of uterocutaneous fistula developed secondary to CS performed 19 years ago.

Baggish et al<sup>6</sup> in 2010 reported another case of uterocutaneous fistula as a complication of ruptured appendix and Crohn's disease during pregnancy.

Pant et al<sup>7</sup> reported in 2012 a case of uterocutaneous fistula of tubercular etiology following CS.

Lim et al<sup>8</sup> 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<sup>9</sup> 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.<sup>10</sup> 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.<sup>10,11</sup>

### 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

- [1] Ruiz-Arteaga JD, Valdez-Murillo AN, Hernández-Trejo MC. Fístulacutáneo-uterina: reporte de un caso y revisión de la bibliografía. *Ginecol Obstet Mex* 2012;80(2):95-98.
- [2] Gupta SK, Shukla VK, Varma DN et al. Uterocutaneous fistula. *Post Grad Med J* 1993; 69(816):822-823.
- [3] Dragoumis K, Mikos T et al. Endometriotic uterocutaneous fistula after cesarean section. *Gynecol Obstet Invest* 2004;57:90-92.
- [4] Promsonthi P, Herabutya Y. Uterocutaneous fistula in term abdominal pregnancy. *Eur J Obstet Gynecol Reprod Biol* 2007;132:239-41.
- [5] Eldem G, Turkbey B, Balas S, Akpınar E. MDCT diagnosis of uterocutaneous fistula. *EJR* 2008;67:e129-e130.
- [6] Baggish M, Bhati A. Uterocutaneous fistula: a complication of ruptured appendix and Crohn's disease during pregnancy. *J Gynecol Surg* 2010;26:163-169.
- [7] Pant PR. Utero-cutaneous fistula: Rare complication of caesarean Section. *NJOG* 2012 Jan-June; 7(3): 66-67.
- [8] Seyhan A, Ata B, Sidal B, Urman B. Medical treatment of uterocutaneous fistula with gonadotropin-releasing hormone agonist administration. *Obstet Gynecol*. 2008; 111(2):526-528.
- [9] Lim PS, Shafiee MN, Ahmad S, Hashim Omar M. Utero-cutaneous fistula after caesarean section secondary to red degeneration of intramural fibroid. *Sex Reprod Healthc*. 2012 Jun;3(2):95-6.
- [10] Burbank F, Hutchins FL Jr. Uterine artery occlusion by embolization or surgery for the treatment of fibroids: A unifying hypothesis—transient uterine ischemia. *J Am Assoc Gynecol Laparosc*. 2000;7:S1-49.
- [11] Sinha R, Sundaram M, Mahajan C. Laparoscopic myomectomy with uterine artery ligation: Review article and comparative analysis. *J Gynecol Endosc Surg*. 2011 Jan-Jun; 2(1): 3-10.

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