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Case Report

Fertility-preserving management of post-adenomyomectomy enterocutaneous fistula with GnRH analogue

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ABSTRACT

Uterocutaneous fistula is a rare pathological communication between the uterine cavity and the anterior abdominal wall, typically presenting with cyclical menstrual discharge through a cutaneous opening. While most cases occur following Caesarean sections or open myomectomy, this case describes an unusual presentation following adenomyomectomy in a 32-year-old nulligravida. The patient initially presented with symptoms suggestive of uterine fibroids but was intraoperatively found to have adenomyosis, for which adenomyomectomy with uterine reconstruction was performed. Four weeks postoperatively, she developed a localized swelling on the lower abdominal wall, which ruptured with the onset of menstruation, discharging blood through both the skin and vagina. Ultrasonography confirmed a hypoechoic tract connecting the uterus to the cutaneous swelling, consistent with uterocutaneous fistula. The patient was successfully managed non-surgically using monthly subcutaneous Goserelin injections for three months, resulting in spontaneous closure of the fistulous tract and resolution of symptoms. This case highlights the diagnostic challenges of differentiating adenomyosis from fibroids using 2D ultrasonography, as well as the importance of considering rare postoperative complications such as uterocutaneous fistula. It underscores the potential role of medical therapy with GnRH analogues as a fertility-preserving alternative to surgery in select cases, contributing to improved clinical outcomes and patient quality of life.

Keywords: Adenomyosis, Adenomyomectomy, Uterocutaneous fistula, GnRH analogue

INTRODUCTION

Uterocutaneous fistula is a pathological connection between the uterine cavity and the overlying skin of the anterior abdominal wall.[1] It is a rare abnormality with only a few cases reported in medical literature. It is clinically characterised by cyclical discharge of menstruum through the cutaneous tract during the menstrual phase of the menstrual cycle, often as a complication of uterine surgery, but may also be associated with endometriosis, pelvic infections such as post-abortion sepsis, uterine malignancies with local infiltration of the skin, prolonged use of abdominal drains, radiation injuries, incomplete closure of uterine incisions, and prolonged retention of foreign bodies such as wound drains.[2,3] The incidence of uterocutaneous fistula is not known with certainty, as the condition is likely to be under-reported in medical literature.

This contributes to the tendency for late diagnosis or outright misdiagnosis, with consequent morbidity to the patient. Reporting such cases is therefore important for improving diagnosis, facilitating counselling, and improving clinical outcomes. Most of the previously reported cases followed Caesarean deliveries, with the remaining occurring after open myomectomy.[4] This case report describes the successful non-surgical management of a 32-year-old nulligravida who developed

uterocutaneous fistula after an adenomyomectomy, thereby emphasizing the importance of considering this condition in postoperative patients with new-onset abdominal wall symptoms.

CASE REPORT

A 32-year-old nulligravida presented to the Gynaecology clinic on account of progressive lower abdominal swelling, dysmenorrhea and menorrhagia of 2 years duration. There was associated passage of blood clots per vaginam, with resultant easy fatiguability and exercise intolerance. She was clinically pale and tachycardic, with a pulse rate of 106 beats per minute. She had a 20-week-size abdominopelvic mass that was not tender, with smooth margins. Other clinical examinations were not remarkable.

A diagnosis of hyaline degeneration of uterine fibroids was made on ultrasonography. She had 3 units of blood transfused on account of haemoglobin concentration of 6.5 mg/dl, and she was counselled for open myomectomy. Intra-operatively, however, there was no fibroid nodule in the uterus. Rather, she had an enlarged, uniformly globular uterus with a consistent texture that suggested adenomyosis.

Adenomyomectomy with uterine reconstruction was performed, and a drain was left in situ, which was removed after 48 hours. She made good clinical recovery and was discharged home on the 4th day after surgery. Adenomyosis was confirmed on histology, and she was discharged from the outpatient clinic. About 4 weeks post-operatively, she presented with a 3.0×2.5 cm well-circumscribed, non-tender swelling that was located about 3 cm superior to the Pfannenstiel incision. The mass ruptured spontaneously during her menstrual flow, which started 3 days later (Figure 1), with concomitant discharge of menstruum through the site and per vaginam. Ultrasonography demonstrated a hypoechoic fistulous tract between the uterus and abdominal swelling (Figure 2).

Figure 1: The cutaneous end of the uterocutaneous fistula, with discharge of menstruum.

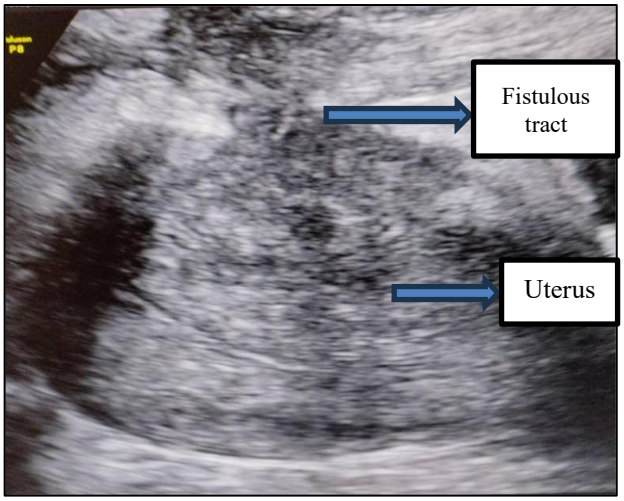

Figure 2: Ultrasonographic image demonstrating the tract of the uterocutaneous fistula.

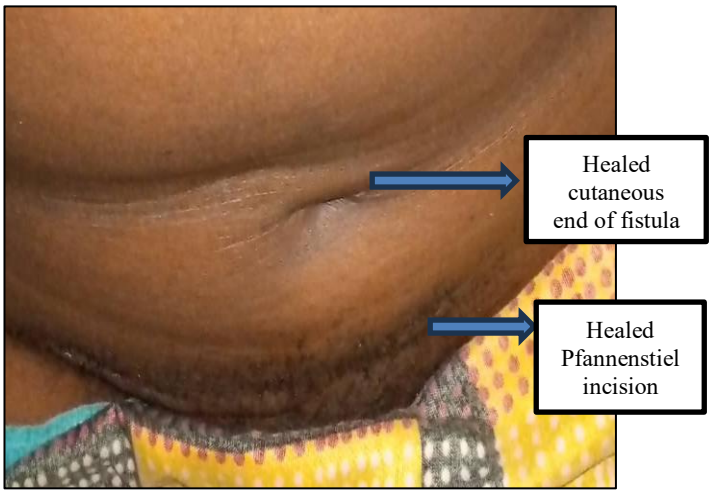

Figure 3: Healed cutaneous end of uterocutaneous fistula post GnRH analogue therapy.

A diagnosis of utero-cutaneous fistula was made, and after due counselling, she was managed with subcutaneous Goserelin, 3.6 mg monthly for 3 months. The menstrual discharge through the anterior abdominal wall ceased, and the fistulous opening closed spontaneously after the 3rd dose of Goserelin (Figure 3). The fistulous tract was also not seen on further assessment with ultrasonography. The patient has remained stable since then, with cyclical menstrual flow.

DISCUSSION

The interrelationship between the clinical features of leiomyoma and adenomyosis continues to pose diagnostic dilemma, as both conditions may present with abdominal swelling, an enlarged uterus, as well as symptoms such as dysmenorrhea and menorrhagia.[5] The dilemma is further compounded when ultrasonography is the only imaging available for evaluation, as in the index case, as both conditions are associated with heterogeneous myometrial

echotexture on ultrasonography. Adenomyosis, however, is typically characterized by diffuse myometrial thickening, antero-posterior wall asymmetry, multiple myometrial cysts, and poorly defined endometrial-myometrial margins.[6] These features, nevertheless, the sensitivity of 2D ultrasonography for the diagnosis of adenomyosis was reported to be 71.1%.[7] The inability to reliably discern adenomyosis from leiomyoma on 2D ultrasonography, particularly in the absence of advanced imaging such as MRI, underscores the need for heightened clinical suspicion and a more holistic diagnostic approach when encountering atypical sonographic or intraoperative findings. Hysterectomy is the surgery of choice for adenomyosis. Adenomyomectomy is, however, a complex and highly technical surgical procedure for women with focal adenomyosis who desire fertility preservation, as in the index patient.[8] The poorly defined endometrial-myometrial junction and the diffuse infiltration of endometrial tissues into the myometrium increase the risk of incomplete removal, extensive myometrial trauma, and impaired integrity of the uterus during the procedure.

Although the clinical outcome of the procedure was initially satisfactory, the patient later developed a utero-cutaneous fistula, which is a serious complication that resulted from a surgical disruption of the poorly defined endometrial-myometrial interface. The utero-cutaneous fistula tract is associated with chronic inflammation and a cyclical response to hormonal fluctuations during the menstrual cycle. This explains the timing of the rupture of the skin swelling and the bloody discharge through the tract during menstruation. Other symptoms may include chronic pelvic pain and purulent discharge from the cutaneous end of the fistulous opening.

Though the diagnosis of uterocutaneous fistula is essentially clinical, imaging modalities such as computerized tomography fistulography, or MRI can provide critical confirmation by delineating the connecting tract between the cutaneous opening and the uterus9. In this patient, 2D ultrasonography proved sufficient to establish the diagnosis, supported by the rapid response to GnRH analogue therapy. Other differential diagnoses of uterocutaneous fistula include rectouterine and vesico-uterine fistulas, which were excluded due to the absence of faecal or urinary incontinence, and the absence of a cutaneous end in these conditions. Scar end endometriosis, though possible, was also excluded in this case, as the cutaneous end of the fistula was separate from the pfannenstiel scar.

The management of utero-cutaneous fistula has conventionally been surgical excision of the tract, with or without hysterectomy.[4,9] However, other modalities of treatment should also consider, based on the clinical manifestation, aetiology and the fertility preferences of the patient. In this instance, the use of Goserelin, which is a gonadotropin-releasing hormone achieved menstrual suppression by inducing hypoestrogenaemia.

Being hormonally responsive, the GnRH analogue achieved complete healing of the fistulous tract within 3 months in this instance, thereby obviating the need for a repeat surgery, especially hysterectomy. It is however noteworthy that medical management may not be successful in women with long-standing, complex or infected fistulas, in which case, surgery remains the hallmark of management, while in other cases, a combination of the 2 modalities of management may be indicated.[10,11]

CONCLUSION

To conclude, this case highlights the diagnostic challenges of differentiating between fibroids and adenomyosis on 2D ultrasonography, with potentially serious implications at surgery. Heightened suspicion is required for the prompt recognition of rare complications of fertility-preserving surgeries for adenomyosis, such as utero-cutaneous fistula. Early diagnosis using imaging, coupled with fertility-preserving hormonal therapy such as GnRH analogues, may obviate the need for repeat surgeries and potential loss of fertility in carefully selected cases.

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