Atypical presentation of vesicouterine fistula (Youssef's syndrome) post-caesarean gossypiboma: a case report

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Abstract

Vesicouterine fistula as described by Youssef manifests with amenorrhea, and cyclic haematuria (menouria) without urinary incontinence commonly following caesarean section. Other aetiologies have been reported for this disease. The post-caesarean section gossypiboma, a dual aetiology causing this condition is rare and can make its presentation atypical, thus posing a diagnostic dilemma. The clinical evaluation alone in the atypical presentation of the vesicouterine fistula is insufficient except complemented by imaging investigations and endoscopic examination of the related organs. We report a case of vesicouterine fistula that was managed initially as chronic pelvic inflammatory disease despite urinary incontinence until further evaluation including a transvaginal ultrasound scan and urethrocystoscopy raised an index of suspicion of a vesical mass suggestive of gossypiboma. The mass was confirmed to be a foreign body, an abdominal gauze pack, or gossypiboma. Its removal and repair of the vesicouterine fistula were associated with a satisfactory outcome.

Introduction

Vesicouterine Fistula (VUF) is an abnormal communication between epithelial linings of the uterus or cervix and bladder.[1,2] It is thought to be rare. However, there have been several reports of this pathology and a recent paper from a West African country reported 36 cases over seven years in a multicentric study.[3] This pathology manifests with constellations of clinical features for which it is termed Youssef's syndrome.[4] The classical description by Youssef in 1957 included amenorrhea, cyclic menouria in the absence of urinary incontinence secondary to a vesicouterine fistula.[4] Depending on the underlying aetiology, this typical description of the symptoms complex is often not evident in some patients yet they may have a vesicouterine fistula. The clinical variability in the presentation of this condition often poses a diagnostic dilemma. As such attending physicians must bear in mind atypical presentations to heighten the index of suspicion of this pathology. Because of this and to put things in a better perspective, Jóźwik and Jóźwik in 2000, came up with their proposition of classification of vesicouterine fistula into three types based on the clinical presentation.[5] Type I has amenorrhea and cyclic menouria without urinary incontinence; type II manifests with cyclic menouria, but has regular menses and urinary incontinence; and type III, which presents with only urinary incontinence, without menouria, and with normal menses.[5] This classification, even though commendable did not accommodate a cohort of patients who may present with urinary incontinence, and amenorrhea without menouria.

Vesicouterine fistula can be said to be uncommon. When compared to other genitourinary fistulae such as vesicovaginal fistula

occurring often from labour-related complications accounting for 71% and 5% for vesicouterine fistula as demonstrated from a study in India.[6] However, with an increasing number of caesarean deliveries being witnessed in most facilities recently, the number of its cases is also on an upward trajectory. It occurs commonly post lower segment caesarean section, especially in repeated cases.[1] Aside from this, other aetiologies also abound and include prolonged labor; use of forceps; normal birth without previous cesarean; operative vaginal birth with vacuum extractor; placenta percreta; manual extraction of the placenta after normal delivery in a woman with previous cesarean; basiotripsy; excision of Gartner cyst; anterior colporrhaphy; endometrial ablation; excision of uterine fibroma after spontaneous necrosis of uterine fibroma; radiotherapy; pelvic trauma; arterial embolization for the treatment of uterine fibroma; invasion of a malignant tumor; tuberculosis; and actinomycosis.[7] There are also reports of this condition after cervical cerclage insertion, secondary to congenital malformations of the genitourinary tract as well as migration of an intrauterine dispositive or foreign body such as gossypiboma.[7]

Gossypiboma is a term used to describe nonabsorbable surgical material such as gauze which is forgotten inadvertently during surgical intervention.[8,9] Though rare, it has been implicated in the causation of patient morbidities and even mortality if not recognized and promptly managed.[10] In a fifteen-year review of 12304 surgical cases managed in southeastern Nigeria, 4 cases accounting for 0.03% were found to have gossypiboma. It has been reported in a patient who had undergone surgery in various parts of the body particularly gynaecological, obstetric, urological, and general surgical interventions.[10,11] Hence, gossypiboma should be a cause for concern to the surgeons and members of the surgical team. It can be a valid reason for medico-legal litigation which may earn the entire surgical team legal punishment.[12] Once a foreign body such as surgical gauze is left intraoperative, two types of pathophysiological processes occur namely exudative reaction and fibrotic reaction resulting in the formation of abscesses, transmural migration with extrusion outside the body on one hand, and adhesions with mass lesions formation on the other hand respectively.[10,12,13,14] The post caesarean section gossypiboma, a dual and rare cause of vesicouterine fistula may make its presentation non-classical or atypical aside from other morbidities in the patient. Morbidities reported to have been associated with gossypiboma include intestinal obstruction, sinus formation, and abdominal abscess or mass among other things.[10,15,16]

The diagnosis of vesicouterine fistula from gossypiboma requires the combined use of clinical evaluation and imaging investigations namely ultrasound scanning, plain radiography, computerized tomography scanning, and magnetic resonance imaging.[15] The diagnostic role of endoscopic examination where applicable, cannot be underestimated. Treatment of gossypiboma can be open or minimally invasive depending on the organ system involved. The most common treatment is surgical, and the routes are: open abdominal, laparoscopic, vaginal, or robotic. However, depending on the available facilities, open surgical exploration is a rational, cheap, and practical approach for patients with gossypiboma-associated vesicouterine fistula. The literature on post-caesarean gossypiboma causing vesicouterine fistula is scarce, let alone the atypical presentation and associated litigation. This forms the basis for this report.

Case Report

A 25-year-old Para 2 + ^{0}, 2 alive, housewife, referred to our facility from a secondary healthcare centre with complaints of abnormal vaginal discharge for 2 years, amenorrhea for 18 months, and lower abdominal swelling for 2 months. She noticed immediately after a second emergency caesarean section 2 years before presentation an abnormal vaginal discharge, which was initially seropurulent, copious, and foul smelling. It later became watery and associated with vulva itching and excoriation of the surrounding skin. She became amenorrhoeic 18 months before presentation after the resumption of her normal menstruation post caesarean section but no associated cyclic haematuria or menouria, antecedent history of menstrual irregularity, or use of contraception. Two months before the presentation, she noticed a progressively increasing lower abdominal swelling with recurrent lower abdominal pain. She had associated painful micturition, anorexia, weight loss, coital withdrawal, and withdrawal from family members. There was no history of fever though had repeated antibiotics treatments at the secondary health facility for recurrent vaginal discharge presumably for pelvic inflammatory disease. There was no associated recent or childhood history of terminal hematuria, necroturia, fecouria, bleeding per vaginam, or history to suggest malignant bowel disease. There was no remarkable comorbidity of note.

Physical examination revealed an acute-on-chronically ill-looking woman with an offensive odour, lethargic, dehydrated, pale, and afebrile (temperature 37.2°C). The respiratory rate was 22 cycles/minute, pulse rate 88 beats/minute, and blood pressure of 110/80 mmHg. There was a midline infra umbilical scar and a palpable suprapubic mass of about 16 weeks gravid uterus size, partly fluctuant, and tender, not attached to the overlying structures. There were no features of peritonitis. The vulva and vagina were smeared with watery, yellowish, foul-smelling discharge. The cervical os was closed. There was a palpable mass in the upper anterior vaginal wall. Digital rectal examination was unremarkable.

The random blood sugar was 6.0mmol/L, complete blood count 5.7×10^{3}/μL with differential granulocytosis of 82.8%, lymphocytes: 34.3%, monocytes - 6.3%, packed cell volume 36.4%, and platelet count 349×10^{3}/μL. The serum electrolytes, urea, and creatinine were normal. Urine was cultured with no bacteria. A transvaginal ultrasound scan (Using Mindray® Ultrasound Scan Machine, DC-30 Model) showed normal ovaries and uterus with an empty endometrial cavity, thickened bladder wall with an intravesical mass measuring 21×23 cm at its upper segment with a calcified margin, and increased vascularity on doppler interrogation (Figure 1).

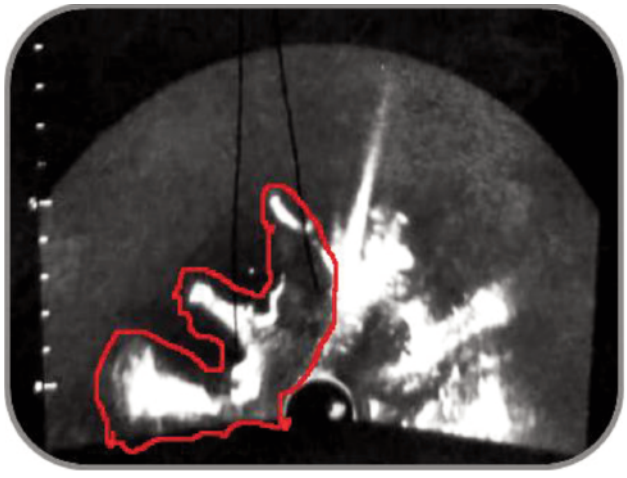

Figure 1. Transvaginal ultrasound scan image showing a urinary bladder mass (circumscribed by red ink) with calcified hyperechoic margins.

She was resuscitated with intravenous normal saline and broad-spectrum antibiotics. Afterward, she had a urethrocystoscopy with findings suggestive of postoperative intravesical foreign body with a differential diagnosis of bladder tumour. She had open bladder exploration, removal of the retained abdominal pack (gossypiboma), and vesicouterine fistula repair (Figure 2). There were findings of dense fibrous adhesion with the retained abdominal pack in the bladder with a calcified portion (Figure 3), vesicouterine fistula with a well-formed and epithelialized tract, and deficient anterior uterine wall. The ureteric orifices were spared and normal. The diagnosis of vesicouterine fistula post-caesarean section gossypiboma was made intraoperative.

She had an uneventful postoperative recovery and was discharged home on a postoperative day 16 for follow-up at the clinic and consultation with the gynaecology clinic for possible contraception.

Discussion

Vesicouterine fistula is a rare pathology that occurs commonly following caesarean section. It is even rarer to have it occurring as a result of post-caesarean gossypiboma. The incidence of gossypiboma is poorly defined in the literature because of underreporting of cases for fear of medicolegal litigation. Notwithstanding, a study from southeastern Nigeria reported an incidence of 0.03%.[10] Several risk factors have been implicated in the causation of vesicouterine fistula but by far the commonest is the post-caesarean section.[1,7] Gossypiboma is another risk factor that can cause this condition but is rare.[10] Both of these risk factors were found in the index patient. The repeat ceaserean section was done as an emergency.

The emergency surgical intervention laid itself to the occurrence of gossypiboma as seen in this case. During emergency surgical intervention, especially when profuse bleeding is encountered, the surgeon as well as his perioperative nurse competence and expertise is put to great task due to increased demand for gauze and abdominal packs to achieve hemostasis.[10] if diligent attention is not given to the gauze and abdominal packs count, any of these can inadvertently be forgotten in situ intraoperative.

Gossypiboma has been reported in a wide range of surgical interventions involving the abdomen, thorax, cranium, and genitourinary organs.[1,7,10] But it is the most common foreign body retained following abdominal surgery which was performed in this patient.[7] The presence of gossypiboma in the body evokes two pathophysiological mechanisms namely the exudative and fibrotic types.[7,13] The exudative type is characterized by suppurative inflammation involving the adjacent visceral wall and in addition to the pressure effect from the gossypiboma, visceral erosion and transmigration may result.[7,13] This process is responsible for the formation of fistulation between two organs as seen in the patient. If the surgical sponge is small, it gets extruded out of the body through the anus, urethra, or vagina.[10,13] The fibrotic type is characterized by the extensive tissue reaction resulting in the formation of adhesions and mass formation.[10] We thought these two pathophysiological mechanisms were at play in the index case. The patient presented with a palpable suprapubic mass and the Intraoperative fistulous tract joining the bladder cavity to the uterine cavity is in keeping with features of fibrotic and exudative mechanisms respectively.

Beyond the pathophysiology, patients with gossypiboma could have symptoms such as abdominal pain, nausea, vomiting, and abdominal mass.[10] Most of these symptoms are also featured in this patient. At times patients with gossypiboma may have mild and vague symptoms which may be ignored as being trivial leading to delayed presentation after months or years of the original surgery. Although the patient presented to our facility two years after the last caesarean section, she made an effort to get her initial mild symptoms that got worse over time treated but were abortive due to a missed diagnosis.

Classically or typically, as described by Youssef, vesicouterine fistula presents with a constellation of features including amenorrhea and cyclic haematuria (menouria) without urinary incontinence.[1,4] The index patient had initial normal menses months post caesarean section but only developed amenorrhea about eighteen months before presentation. The absence of cyclical haematuria and urinary incontinence makes this presentation non-classical or atypical. The implication is the diagnostic dilemma that can arise during evaluation and which this patient suffered. The dual aetiology of vesicouterine fistula namely repeated ceaserean section and gossypiboma might be the reason for the diagnostic challenge. It

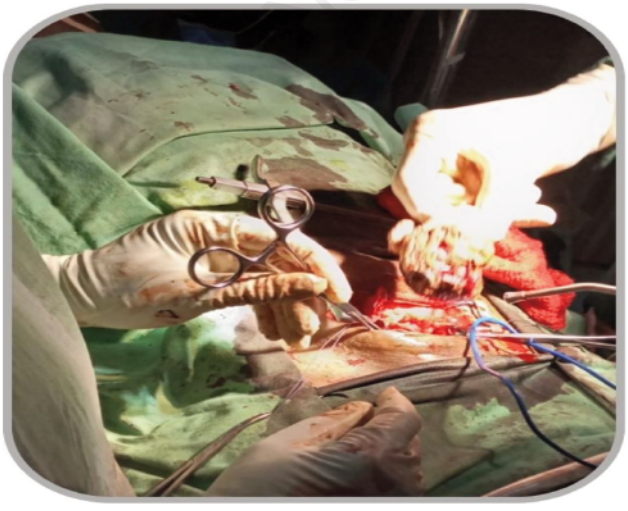

Figure 2. Abdominal pack being removed from the bladder.

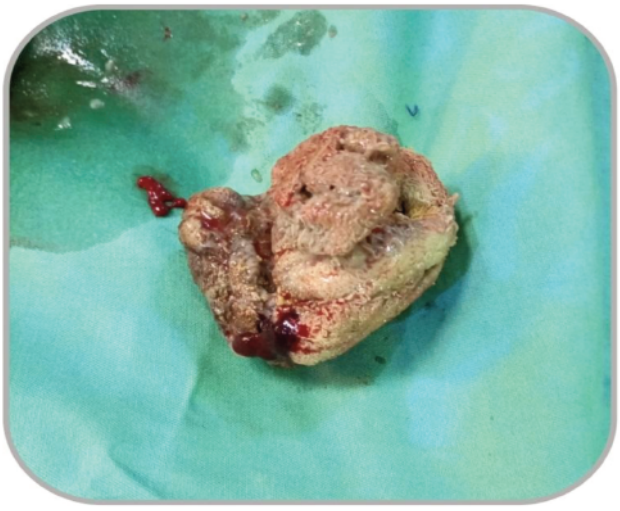

Figure 3. Abdominal pack being removed from the bladder.

calls for a high index of suspicion of vesicouterine fistula and the need for further investigations like imaging with persistent patient symptoms despite repeated treatment for chronic pelvic inflammatory disease.

The diagnosis of vesicouterine fistula post ceaserean gossypiboma can be tasking. The task can be made easy by combining the role of clinical evaluation, imaging investigations, and endoscopic examination.[15,16] The regional ultrasound scan, plain radiograph, computerized tomography scan, and magnetic resonance imaging when available are useful and can complement clinical evaluation in the diagnosis. The regional transvaginal ultrasound scan and urethrocystoscopic examination performed on the patient greatly enhanced her diagnosis and decision for definitive intervention.

Conclusions

The vesicouterine fistula post caesarean gossypiboma is an uncommon entity in the literature. As more rare aetiology of the vesicouterine fistula are discovered, so shall be encountered the atypical presentation and the diagnostic dilemma of this pathology. Therefore, emphasis on a high index of suspicion with the combination of clinical evaluation, imaging investigations, and endoscopic examination is vital to avoid missing the diagnosis. Open surgical intervention is a good option in its management and is associated with a favourable outcome. This notwithstanding, post-caesarean gossypiboma as a cause of vesicouterine fistula is better eliminated or prevented at all costs because of the risk of medicolegal litigation.

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