

# Incidentally diagnosed post-cesarean vesicouterine fistula (Youssef's syndrome)

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## Abstract

Vesicouterine fistula (VUF) is a very rare occurrence and is estimated to occur in only 1–4% of all genitourinary fistulas; 90% of cases are Youssef syndrome, which is accompanied by amenorrhea and cyclic hematuria (menouria). In this article, a renal transplant donor who was incidentally diagnosed with Youssef syndrome 20 years after a second cesarean delivery.

## Introduction

Youssef syndrome was defined in 1957 and it comprises of a history of previous cesarean delivery, amenorrhea, and cyclic hematuria.<sup>1</sup> It is a rare complication due to accidental opening of the bladder during cesarean delivery.<sup>1,2</sup> In the VUF that occurs in 1–4% of all urogenital fistulas, most patients do not experience any urinary incontinence.<sup>2–4</sup> Although VUF can be seen after primary cesarean delivery, it is most commonly seen after multiple cesarean sections and incidence tends to increase with incremental use of low-segment cesarean section.<sup>5–7</sup> In this report, we present a VUF case incidentally detected 20 years after second cesarean section.

## Case report

Ovarian cyst was detected in routine examination of a 49-year-old woman after she became a kidney donor candidate for her husband and was referred to the gynecology outpatient clinic before transplantation. In the anamnesis taken by the gynecologist, it was detected that the patient, who had a history of two low-segment cesarean sections in 1984 and 1989, had amenorrhea and described cyclic hematuria over a period of nearly 20 years. The patient,

whose computed tomography (CT) urography had no pathology, was referred to us for further examination of hematuria.

In the voiding cystourethrography of the patient (Figs. 1 and 2), it was detected that the contrast agent given to the bladder was passed to the uterine cavity. It was thought that cyclic hematuria of the patient was menouria and the patient was diagnosed with VUF (Youssef syndrome) seen after cesarean section. Total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH-BSO) operation was recommended by the gynecology clinic.

## Discussion

VUF is a rare pathology seen especially after cesarean sections.<sup>8</sup> VUF was previously seen after vaginal delivery performed with forceps, but now 83–93 % of cases are seen after cesarean section.<sup>9</sup> Some risk factors for VUF include: insufficient dissection of the bladder from the lower segment of the uterus, excessive intraoperative bleeding, use of vacuum and forceps, manual removal of the placenta, placenta percreta, uterus rupture, previous cesarean section, and abortion history. Other less common causes are endometriosis, inflammatory bowel diseases, migration of intra-uterine device, bladder tuberculosis, and congenital lesions.<sup>10,11</sup>

Jozwik et al classified VUF cases according to menstruation status in 2000.<sup>12</sup> In Type 1, there is amenorrhea with menouria (cyclic hematuria) and it is named as Youssef syndrome. In Type 2, there is hypomenorrhea and menouria. In Type 3, menarche status of patient is completely normal and patient has no complaint of menouria.<sup>12</sup> Most cases (90%) have Type 1 classification.<sup>13</sup> Our patient belongs to Type 1 VUF classification.

Youssef syndrome seen after cesarean section is characterized by amenorrhea accompanied by menouria, lack of urine incontinence, vesicouterine fistula, and normal cervical canal.<sup>14</sup> Although complaints are often due to amenorrhea and cyclic hematuria, they can also be related to



**Fig. 1.** Voiding cystourethrogram of the patient.

urinary incontinence or infertility.<sup>15</sup> Interestingly, although our patient had these symptoms, she has lived with them for almost 20 years without seeing a physician. In the gynecological evaluation of the patient due to ovarian cyst detected during routine controls performed for being a renal transplant donor, fistulas were suspected and incidentally detected. As far as we know, this is the first report in the literature of incidental detection of fistulas.

To demonstrate the fistula, voiding cystourethrogram and cystoscopy are not always sufficient and it was found in a study that pelvic magnetic resonance imaging (MRI) showed 100% diagnostic accuracy.<sup>16,17</sup> In cystoscopy, the fistula opening is always in the supratrigonal region.<sup>18</sup> Cystoscopy was not performed in our case and voiding cystourethrogram was sufficient for diagnosis.

For small fistulas detected early, a conservative treatment approach is preferred, as spontaneous recovery has been reported to occur in 5% of cases.<sup>3,19</sup> Urinary infection is treated with a conservative approach and the patient is followed up for three weeks via catheterizing the bladder. Surgical approach is performed for all wide fistulas and small fistulas that do not have spontaneous recovery after a three-month conservative followup.<sup>20,21</sup> For a surgical approach, options include trans-abdominal, endoscopic, and robotic procedures; hysterectomy is also an option if there is no desire for fertility.<sup>22</sup> If there is desire for fertility, fistula repair is essential and success rate is high; pregnancy after repair has been reported.<sup>2,23</sup>

Due to lack of desire for fertility in our case, TAH-BSO operation was performed by the gynecology team and bladder wall repair was preoperatively performed by us.



**Fig. 2.** Voiding post-voiding film of the patient.

## Conclusion

VUF diagnosis is not difficult; it is often accomplished by a combination of symptoms and patient history. Treatment is varied and includes conservative, medical, or surgical options. Total abdominal hysterectomy is advisable in patients for whom fertility is not desired.

**Competing interests:** The authors declare no competing financial or personal interests.

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