

## PREGNANCY FOLLOWING SPONTANEOUS CLOSURE OF A VESICOUTERINE FISTULA

Mehmet EKİNCİ, Mustafa Burak HOŞCAN, Ahmet TUNÇKIRAN

Başkent Univesity, Department of Urology, Alanya Training and Reseach Center, ANTALYA, TURKEY

### ABSTRACT

**Introduction:** Vesicouterine fistula without vaginal leakage of urine, cyclic hematuria and amenorrhea is a rare condition, referred to as Youssef's syndrome. It is a rare complication of caesarean section when bladder injury occurs and a fistula develops. Although standard treatment of this syndrome is surgical repair, we suggest that surgical repair is not always necessary, and that recovery even pregnancy may follow conservative management and spontaneous healing.

**Key words:** Cesarean section, Uterovesical fistula, Youssef's syndrome

### ÖZET

Vajinal idrar kaçağı olmadan vezikouterin fistül, siklik hematüri ve amenore birlikteliği Youssef sendromu olarak adlandırılan ender bir durumdur. Sezaryen ameliyatının ender bir istenmeyen yan etkisi olan bu klinik tablo, mesane yaralanmasına bağlı fistül geliştiğinde görülür. Bu durumun standart tedavisi cerrahi onarım olsa da, koruyucu tedavi ve kendiliğinden iyileşmeyle de düzelme ve hatta gebeliğin görülebileceğini bildirmektediriz.

**Anahtar kelimeler:** Sezaryen, Uterovezikal fistül, Youssef sendromu

### INTRODUCTION

In 1957, Youssef described the classic triad of caesarean section, amenorrhoea, and cyclic haematuria (menouria) in the absence of urinary incontinence as a syndrome, which is characteristic of vesicouterine fistula<sup>1</sup>. Youssef syndrome is a rare complication of caesarean section following inadvertent bladder injury. Vesicouterine fistula is one of the least common types of urogenital fistula, accounting for only 1-4% of all cases<sup>2</sup>. Commonly, in modern obstetric practice, vesicouterine fistulas follow the lower segment type of caesarean section which accounts for 83% of cases<sup>2</sup>. Rarely, vesicouterine fistulas follow long labor, forceps delivery, vaginal birth after caesarean section, abdominal pregnancy for perforation of the anterior wall of the uterus, gynaecological injuries, tuberculosis of the genital tract, or intrauterine contraceptive devices<sup>3-5</sup>. Improved obstetrical and surgical practice is responsible for the decrease in the incidence of these fistulae in comparison with a much more frequent occurrence in the 19th century, but the prevalence worldwide is now increasing because more caesarean sections are being carried out. It is important to be aware of its clinical presentation as well as the potential for non-surgical management.

### CASE REPORT

A 28-year-old female, who already had had 2 deliveries by caesarean section, was referred to our

unit with vaginal urinary leakage, which developed two months after her third delivery that was performed by caesarean section. The urinary leakage was occurring only when the bladder was over distended. She was continent for the most of time. Patient was evaluated by clinical history and physical examination. Blue methylene test, intravenous urography with cystogram phase, transvaginal ultrasonography, pelvic tomography, cystoscopy, vaginoscopy and hysteroqram was also carried out. Blue methylene test was positive. In intravenous pyelogram endometrial cavity was opacified in 15 minutes (Figure 1). Contrast material was seen at endometrial and vaginal cavity in uterine slices of pelvic tomography (Figure 2). The passage of contrast material from approximately 5 mm diameter fistula tract into the bladder was seen on hysteroqram (Figure 3). Consequently the patient was admitted for examination under anaesthetic and cystoscopy showed a fistulous track between the anterior isthmus of the uterus and the posterior wall of the bladder. It was decided a conservative treatment by draining the bladder by a transurethral Foley catheter which was left indwelling for. Two months later the patient recovered her menses, which presented with cyclic bloody urine from the catheter and amenorrhoea. Unfortunately, spontaneous closure of the fistulous track did not occur. After 4 months the patient still complained total vaginal urinary leakage as well as menouria

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**Figure 1.** Passage of contrast material from bladder into the endometrial cavity in intravenous pyelogram



**Figure 2.** Contrast material seen at endometrial and vaginal cavity in uterine slices of pelvic tomography.



**Figure 3.** Passage of contrast material to bladder in hystero-gram

with amenorrhoea. Cystography still showed the abnormal vesicouterine fistulous communication. The fistulous tract was thoroughly fulgurated by

endoscopically. Foley catheter drainage is left indwelling for 6 weeks. After 6 weeks the catheter was removed and the patient was totally continent but cyclic menouria continued. Thus it was decided to surgical repair of the fistula. But the patient refused the surgical repair and run out of our follow-up. Surprisingly, after two and a half year the patient admitted to our clinic again with a 5 months pregnancy. In clinical history, she told that the menouria was continued for one and a half year and she had no symptoms for last one year. She had an uneventful follow-up for the pregnancy and gave birth to a child without complication.

## DISCUSSION

Vesicouterine fistulas may develop just after a caesarean section or in the later puerperium as well as occur after repeated procedures<sup>5,6</sup>. While performing a caesarean section the bladder may be damaged by direct injury, inadequate downward mobilization or aberrant sutures. Delayed vesicouterine fistula formation may result from infection, devascularization, clamping or haematoma of the urinary bladder<sup>2</sup>. The fistulous communication is usually reported between the posterior supratrigonal part of the bladder and the anterior lower segment of the uterus or, more rarely, the cervix. Youssef proposed that the syndrome develops if the fistula is above the level of the uterine isthmus, which acts as a 'sphincter'. When an uterovesical fistula is below this level, however, menstruation occurs normally but urinary incontinence occurs.

Spontaneous healing is reported in 5% of cases by conservative approach<sup>7</sup>. Conservative management includes bladder catheterisation for at least 4-8 weeks, and hormonal management by oestrogen and progesterone-induced amenorrhoea<sup>7,8</sup>. This conservative management is particularly successful when the fistula is discovered early. Endoscopic fulguration is effective for selected, small fistulae<sup>9</sup>. Surgical repair should be deferred for at least 2-3 months after caesarean section to allow complete uterine involution and resolution of inflammation. Effective and successful treatment of vesicouterine fistulas is followed by disappearance of vaginal leakage and/or menouria with the recovery of normal menses. The pregnancy rate after repair of vesicouterine fistulas is reported as 31.25 % with a rate of term deliveries 25%<sup>10</sup>.

When Youssef first described this syndrome in 1957, he pleaded ‘that every case of this nature should be fully investigated and reported, . . . it is only in this way that the pathogenesis of its unique features can be fully understood’<sup>1</sup>. Although preferred treatment of this syndrome is surgical repair, several different approaches have been advocated for the treatment of vesicouterine fistulas. Spontaneous resolution may occur. We also suggest that surgical repair is not always necessary and that recovery even pregnancy may follow conservative management and spontaneous healing.

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