Endometriosis arises from implant of endometriotic cells outside the uterus: A report of active vesicouterine centrifugal fistula

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Summary

Vesicouterine fistula is rare, accounting for nearly 4% of all urogenital fistulas. Lower segment cesarean delivery is the main predisposing event but in the last few years other possible predisposing factors have been pointed out. Clinically, it can show itself in different forms and the diagnosis is often delayed although it is not difficult.

We report our experience about a case of postcesarean vesicouterine fistula arising on a focus of vesical endometriosis and we discuss an eventual hypothetical pathogenetic correlation between bladder endometriosis and uterovesical fistula.

Key words: Bladder endometriosis; Vesicouterine fistula; Obstetric surgery.

Introduction

The incidence of urinary tract lesions in gynecologic and obstetric surgery is relatively insignificant both because of improvements in several surgical techniques and because in most cases the lesion is located and adequately repaired during surgery.

Vesicouterine fistula is a rare entity, accounting for approximately 4% of all urogenital fistulas [1], and is often the result of a cesarean delivery. The lesion can occur when, owing to a unsuitable maneuver, during separation between the bladder and lower uterine segment, the vesical wall is injured and the lesion ignored.

A case of postcesarean vesicouterine fistula, arising on a focus of vesical endometriosis occurring in our Department is reported.

Case

G. P., was a 36-year-old patient with obstetric anamnesis of two spontaneous abortions and two pregnancies obtained with cesarean delivery, the last of which was five months before. During the last cesarean delivery, performed in another hospital, she incurred a vesical lesion which was intraoperatively repaired.

Because of persistent cyclical hematuria, together with absence of menstrual flow out of the vagina, she was hospitalized to ascertain if the diagnosis was a suspected vesicouterine fistula. A cystographic X-ray test revealed a diverticular formation of the vesical dome, resulting from surgery and that finding was successively confirmed by urography with perfusion.

Transurethral cystoscopy revealed a diverticular formation with a neck unsurmountable by cystoscope and with easily bleeding internal walls, particularly on the posterior wall.

The patient underwent transabdominal surgical reconstruction of the lesion in order to remove the uterovesical fistulous duct.

Results

The operation was performed anatomically dissecting and dividing the bladder from the uterus at the level of the fistula, identifying the limbi and repairing each organon separately by absorbable ligatures. During the postoperative period vesical decompression was performed by means of a catheter for ten days, associating the medical therapy of antibiotics and anti-inflammatories with the complete restitutio ad integrum of the patient.

Histologic examination of the uterovesical fistulous duct stab macroscopically demonstrated a fibrose flap, partly invested with mucosa and about 2 cm in maximum diameter, in which there was cleavage with a bleeding wall along the thickness of the examined piece. Microscopic description showed a flap of vesical wall, partly covered by urothelium, in which numerous glands covered by cylindrical simple epithelium were observed. This epithelium had dilated lumen and recent and previous hematic intraluminal infiltrations. The description was indicative of vesical endometriosis.

The patient's check-up six months after surgery and Gn-RH analogue therapy confirmed complete remission of symptomatology and the patient was considered healthy.

Discussion

Vesicouterine fistulas are rare disorders among the lesions of the urogenital system, but in the last few years the cases reported in the literature have become more and more frequent [2-5], better defining the diagnostic and therapeutic etiologic aspects of this pathology.

All authors agree about the fact that the most common event associated with vesicouterine fistula is a previous lower segment cesarean delivery [2-4, 6, 7] which is due to the changes in obstetric practice during the last half of the 20th century and mostly to the increase in the incidence of cesarean deliveries.

Revised manuscript accepted for publication December 21, 2000

Generally the lesion occurs during the preparation of the anterior peritoneal (vesicouterine) flap, or when the bladder is displaced because of previous operations or pathologic causes which caused large adhesions or infiltrated the vesical musculature as in cases of neoplastic or endometriosic involvement.

However an accurate analysis of the existing literature also shows other probable predisposing events, such as previous brachytherapy [8], use of intrauterine contraceptive devices [9], uterine biopsy [10], previous abdominal hysterectomies [11] or vigorous dilatation of the cervix during labour [2].

As well as in our report, clinically in most cases the typical symptomatology consists of cyclical hematuria (menouria), but also other kinds of clinical presentation are observable, such as intermittent vaginal urinary leakage or mixed forms with more or less regular cyclical menses [2, 12].

In 1998 Jozwik [13] suggested a new clinical classification of the disease, based on menstrual flow: type 1 with menouria, type 2 with double flow both through the vagina and the bladder, or type 3 with normal vaginal menstruation.

Generally the above described symptomatology appears several months after surgery, until lactation amenorrhea ceases, and clearly it is the cause of delayed diagnosis which is relatively easy by cystoscopy and cystography.

However, in those rare cases in which conventional radiological and endoscopic techniques are not determinant for the diagnosis, heavily T-2-weighted-MRI can be used successfully to demonstrate fluid within the fistula [14].

It is clear how a correct diagnosis, possibly precocious, can induce optimal therapy, which in most cases is surgical since the fistula is usually small and easily accessible abdominally. However some cases have been successfully treated laparoscopically or with spontaneous closure after medical treatment [15, 16].

Physiopathologically some authors [12, 17] report how leakage of urine into the vagina could be prevented by the action of the cervical isthmus as a sphincter and this could explain the sometimes intermittent nature of leakage. The condition was thought possibly to be pressure-related inasmuch as urine could leak through the fistula in patients with elevated bladder pressure or with detrusor hyperreflexia. We do not agree completely with the hypothesis described because it seems highly unlikely that the cervical os is a sufficient barrier to prevent urine leakage but more studies are needed to better understand the pathophysiology of the lesion.

Jozwick *et al.* [5] suggest moreover a possible role of estrogens and of endometrium in the formation of the fistula, affirming that at least some of the vesicouterine fistulas follow the criteria of external endometriosis. Hypothetically we agree with this assessment, both on the basis of histopathological findings revealed in our case and on the basis of recent studies suggesting that deep endometriosis could be a consequence of external adenomiosis [18, 19].

However, in consideration of the scarce information in the literature, at the present time it is not possible to say this hypothesis is valid. In the meanwhile, depending on the prevalence of endometriosis, we could suggest a new classification based on two entities: 1) a classic fistula with urine leakage and 2) a transmural adenomyotic lesion, which is normally closed and only bleeds during menstruation into the uterus or into the bladder depending on the lesion.

In our analysis only two cases were found to have some correlations between the presence of urogenital fistulas and endometriosis: Mercader *et al.* [10] report a case of a patient with adenomyosis and pelvic adhesions, who presented menouria after uterine biopsy, while Yazawa *et al.* [20] report a case of vesicoadnexal fistula following endometriosis of an ovary. Certainly a vesical endometriosis, primary or secondary to the spread of an endometriosis of the vesico-vaginal septum can involve vesical mucosa, causing typical symptomatology. Therefore the following phlogistic state could be hypothesized as having an eventual role in the pathogenesis of fistulous vesicogenital stabs, also considering that the finding of vesical endometriosis is not rare in patients who have undergone previous gynecological operations [21, 22].

In conclusion there is relatively little information about the pathophysiology of the lesion, a consequence of its relative rarity. Other studies are needed to understand, prevent and adequately treat this clinical disorder, but it is prudent to hypothesize potential endometriosis when even few endometriotic cells accidentally adhere to non endometrial tissue.

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