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CASE REPORT

Cesarean scar caseating granuloma: a case of vesicouterine fistula 30 years after cesarean section

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Key Clinical Message

A mass developing in operating scar part with fistula should raise concern for caseating granuloma even if many years after operation.

Keywords

Caseating granuloma, cesarean scar, PCR for mycobacterium, vesicouterine fistula.

Introduction

The most common cause of a vesicouterine fistula is lower segment cesarean section [1]. When present, a vesicouterine fistula usually occurs within a few months postcesarean section, although subclinical a vesicovaginal fistula can also occur in patients with a history of genital tuberculosis [2].

We describe a case in which cesarean scar infection developed into a uterine mass and vesicouterine fistula 30 years postcesarean section.

Case History

A 69-year-old woman presented at our hospital with a complaint of genital bleeding that had persisted for a few days. She reported a history of ectopic pregnancy at 26 years of age, cesarean section at 29 years of age, and menopause at 53 years of age. She also had several uterine myomas, which were discovered on gynecological examination performed 6 months previously. She denied any fever, cough, abdominal pain, or gross hematuria.

On examination, she exhibited normal uterine size without tenderness. Transvaginal sonography revealed a hypoechoic mass in the lower uterine segment (Fig. 1). Pelvic MRI revealed a vesicouterine fistula through the mass (Fig. 2). Blood tests showed normal results, with the exception of mild neutrophilic (75%) leukocytosis (11,880 cell/mm^3) and anemia (Hb 10.2 g/dL). Tumor marker determination (CA125, CEA, CA 19.9) showed negative results. Microscopic examination of urinary sediment revealed pyuria (100 white blood cells/high-power field (hpf)) but no hematuria (1–4 red blood cells/hpf).

The patient was referred for investigation to exclude malignancy, and an endometrial biopsy was performed. Histological examination revealed granulation tissue containing lymphocytes, neutrophils, and plasma cells, with chronic inflammation, calcification, and necrosis. Repeat biopsy confirmed the presence of granuloma without malignancy. Staining for mycobacteria and fungi yielded negative results. The patient subsequently underwent cystoscopy, which revealed a mass protruding from the posterior wall of the bladder, covered with normal urothelium.

Laparotomy was performed for further histological evaluation and repair of the vesicouterine fistula. Intraoperative exploration exposed a purulent-like mass at the

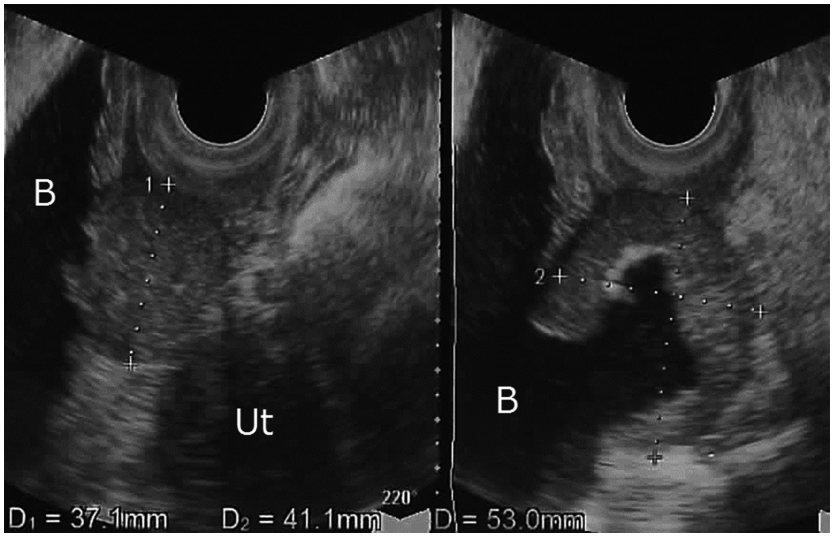

Figure 1. Pelvic ultrasound exhibiting a homogenous isoechoic solid mass (5.3 × 4.1 × 3.7 cm) growing through the lower uterine segment into the bladder. Ut: uterus, B: bladder.

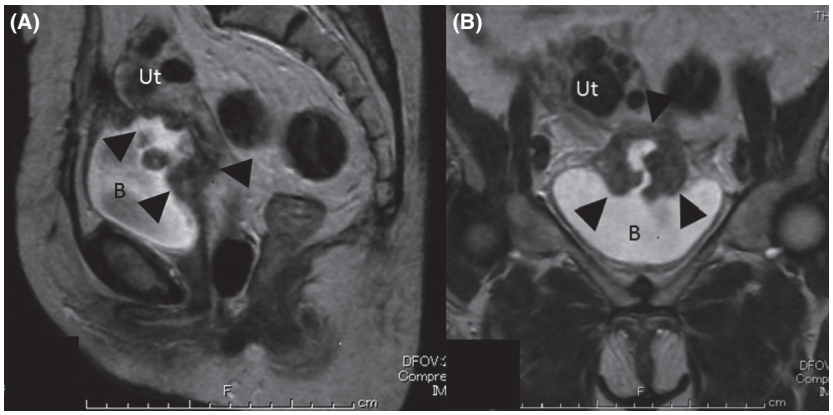

Figure 2. T2-weighted MR images revealing uterine mass with vesicouterine fistula (arrow heads). (A) T2-weighted sagittal MR image. (B) T2-weighted coronal MR image. Ut: uterus, B: bladder.

lower uterine segment with bladder wall invasion. Total hysterectomy, bilateral oophorectomy, partial cystectomy, and ureterocystoneostomy were performed (Fig. 3). A yellowish white sarcoid measuring 4.7 × 4.5 × 4.5 cm between the bladder and the anterior wall of the uterus was macroscopically visualized. Histological examination of the surgical specimen revealed a caseating granuloma (Fig. 4). Ziehl–Neelsen, as well as Periodic acid–Schiff (PAS) staining yielded negative results.

Following the surgery, we performed several examinations to determine the cause of the caseating granuloma. Chest computed tomography revealed normal results. No mycobacteria or fungi could be cultured from the operative specimen, urine samples, or sputum sample and PCR assays for *Mycobacterium tuberculosis* DNA showed negative results. Nested PCR assays for *M. tuberculosis* DNA, as described by Azov et al. [3], also showed negative results. Syphilis, another cause of caseating granuloma, was excluded by RPR and TPHA screening. The patient has remained free of recurrence for 60 months, without antimicrobial therapy.

Discussion

In this patient, cesarean scar infection led to the development of a uterine mass and vesicouterine fistula 30 years postcesarean section. Caseating granuloma is typically caused by tuberculosis, atypical mycobacterial infection, syphilis, or certain species of coccal fungi, including those in cryptococcosis and coccidioidomycosis.

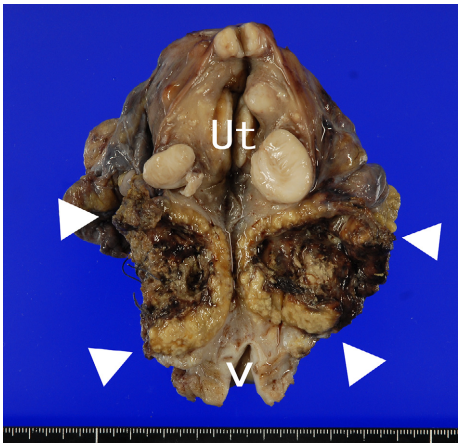

Figure 3. Bisected hysterectomy specimen revealing a yellowish 4 cm mass with central necrosis arising in the lower uterine segment. V: vagina, Ut: uterus.

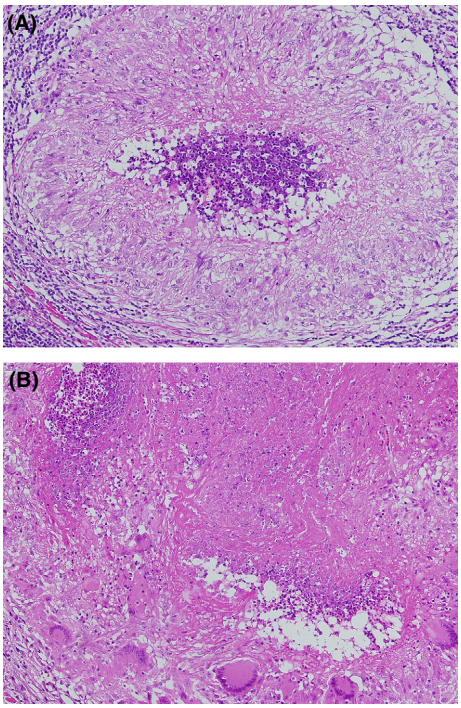

Figure 4. Hematoxylin and eosin stains at ×400 magnification. (A) Central necrosis surrounded by palisading epithelioid cells. (B) Langhans' giant cells surrounding an area of caseous necrosis.

In this case, although Ziehl–Neelsen and PAS staining yielded negative results, atypical mycobacterial or cryptococcus infections were considered as possible causes when the method of elimination was applied. Considering the nested PCR assay for *M. tuberculosis* described by Park et al. [4] using DNA extracted from a well-formed granuloma with caseous necrosis and Langhans-type cells was positive for all of the 13 tuberculosis cases tested in their study, our negative result on nested PCR for *M. tuberculosis* using DNA extracted from the caseating granuloma could rule out tuberculosis. Negative TPHA results rule out syphilis. If the pathogen is confirmed, further PCR assays for atypical mycobacterium and fungus are needed.

Ascending infection from endometritis was also considered as a route of myometrium infection in the absence of pulmonary lesions. Mycobacterium or fungi could hypothetically infect the uterine myometrium following cesarean section. However, endometritis rarely leads to abscess formation or granulation because of the excretion of fungal bodies from the uterus. Therefore, we speculated that a caseating granuloma developed in the myometrium as a result of cesarean scar infection.

A vesicouterine fistula usually occurs a few months after cesarean section [5]. However, in some cases, vesicouterine fistulas have been known to develop months or even 30 years later [6]. Thus, we believe that an indiscernible infection may have occurred in the patient's youth and might have become apparent after menopause with the development of the mass and the vesicouterine fistula. In addition, the fact that primary mycobacterial endometritis is rare in postmenopausal women [7,8] also supports the possibility that the infection occurred at a younger age, not after menopause.

Based on the foregoing, we think it possible that earlier unidentified cesarean scar infection with mycobacterium or fungi could be revealed by the presence of a vesicouterine fistula 30 years following cesarean section.

Secondly, if endometrial biopsy of the low echoic myometrial mass reveals granulation tissue instead of malignancy, total hysterectomy is recommended for the diagnosis and treatment in consideration of the caseating granuloma.

There are few examples of myometrial tuberculosis on ultrasound image. Ultrasound of this patient revealed a low echoic homogenous mass with a relatively clear border. This resembles ultrasound images of tuberculosis of the breast and lymph nodes [9]. MRI scans closely resembled leiomyoma, with increased signal intensity on T1-weighted images and slightly lower signal intensity on T2-weighted images. In the absence of a defined set of characteristics for tuberculosis scar formation on ultrasound and MRI images, differentiation from other uterine tumors such as denaturing uterine myoma or uterine cancer is necessary. In this case, biopsy of the myometrial mass revealed granulation tissue, but was not able to reveal the caseating necrosis. However, since total hysterectomy is recommended for treatment of a vesicouterine fistula [5], total hysterectomy is also thought to be necessary for the diagnosis and treatment of cesarean scar granuloma with a vesicouterine fistula.

Conclusion

We should consider caseating granuloma as a differential diagnosis for tumors since it can develop in the surgical scar many years later.

Conflict of Interest

None declared.

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