

Case Report

Ulcerative interstitial cystitis in an adolescent successfully treated with complete transurethral ulcer resection: A case report

Eri Yuki,  Kenji Obara, Hiroo Kuroki, Hiroyuki Yamazaki, Kazutoshi Yamana, 
Akira Tadokoro, Kaede Hiruma and Yoshihiko Tomita

Division of Urology, Niigata University Graduate School of Medical and Dental Sciences, Niigata, Japan

Abbreviations & Acronyms

DMSO = dimethyl sulfoxide
OSPI = O'Leary-Sant
problem index
OSSI = O'Leary-Sant
symptom index
PI = problem index
PUF = pelvic pain and
urgency/frequency patient
symptom scale
SI = symptom index

Correspondence: Eri Yuki
M.D., Division of Urology,
Niigata University Graduate
School of Medical and Dental
Sciences, Asahimachi 1-757,
Chuo-ku, Niigata 951-8510,
Japan. Email: eyukiey-
niimed@yahoo.co.jp

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Introduction: Interstitial cystitis is difficult to treat and may affect adolescents.

Case presentation: A 15-year-old girl presented with severe pain upon terminal micturition that persisted for approximately 2 hours. The pain had been present for more than 1 month. Cystoscopy revealed severe erosion throughout the trigone. Transurethral fulguration did not improve her symptoms. However, complete electric resection of the ulcer markedly reduced the symptom. After complete resection, pain on urination disappeared and she has had no pain without medication for 15 months.

Conclusion: Complete resection not fulguration of the ulcer is effective for interstitial cystitis in adolescent female patients.

Key words: adolescent, complete transurethral resection, ulcerative interstitial cystitis.

Keynote message

For severe ulcerative interstitial cystitis in adolescents, complete electric resection not fulguration may lead to remarkable effect.

Introduction

Interstitial cystitis is difficult to treat and may affect adolescents.¹ It is commonly categorized into ulcerative or non-ulcerative forms based on cystoscopy. The ulcerative form, such as Hunner ulcers, is associated with more severe pain. We herein report a case of ulcerative interstitial cystitis in an adolescent who was successfully treated by complete transurethral resection of the ulcer.

Case presentation

A 15-year-old girl presented with severe pain upon terminal micturition, a pain that persisted for approximately 2 hours. When pain occurred, she was unable to move. The pain had been present for more than 1 month. She was treated with multiple antimicrobials, but no improvement was observed. Although urinalysis revealed a high white blood cell count, urine culture, cytology, polymerase chain reaction for tuberculosis, and adenovirus infection were all negative. The blood interferon-gamma assay for tuberculosis infection was also negative. She underwent cystoscopy, hydro-distention, and bladder biopsy under general anesthesia. Cystoscopy demonstrated severe erosion throughout the trigone (Fig. 1). Post-distension capillary hemorrhage was observed in only a small part of the posterior wall. Her bladder capacity was 700 mL at 80 cm H₂O. Electric fulguration was performed for the trigonal ulcer. Severe inflammation with granulation and lymphocyte infiltration were found on histological assessment of the bladder biopsy from the trigone (Fig. 2). Postoperatively, her symptom improved temporarily, but gradually returned to the preoperative status 1 month later. Although several medications were tried in combination, including suplatast tosilate at 100 mg TID, tranexamic acid at 250 mg TID, potassium citrate/sodium citrate hydrate at 1 g TID, and loxoprofen sodium hydrate at 60 mg TID, her symptoms did not improve. Two months after the first

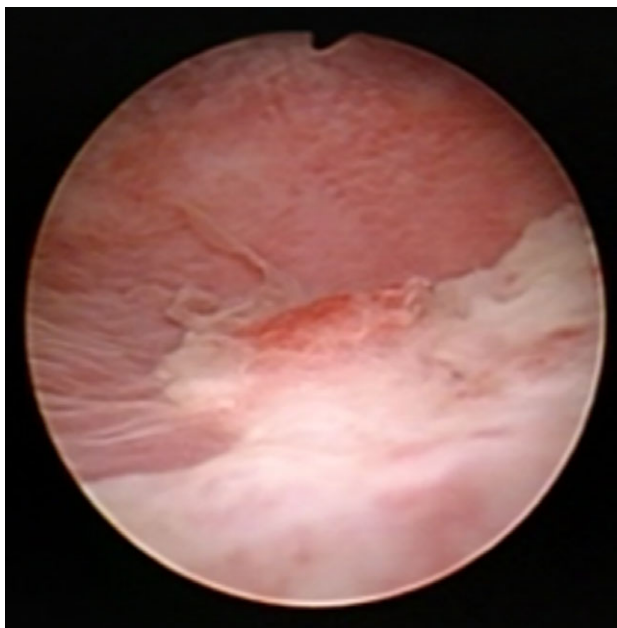


Fig. 1 Cystoscopy revealed severe erosion throughout the trigone at the first operation.

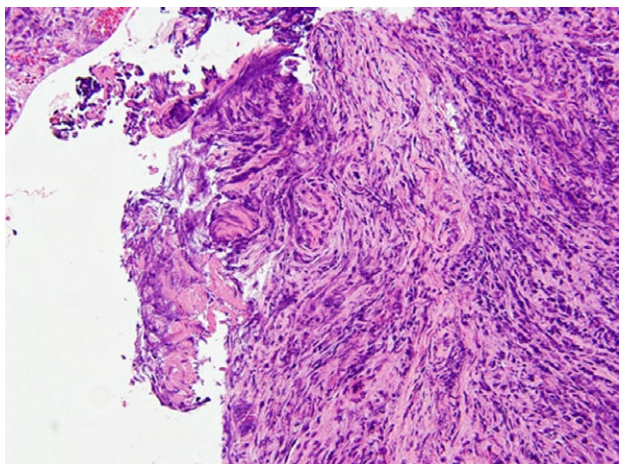


Fig. 2 Histological assessment of the bladder biopsy from the trigone demonstrated severe inflammation with granulation and lymphocyte infiltration.

Table 1 Changes in clinical symptom scales

	Before DMSO	After DMSO	Before resection	After resection
OSSI (0–20)	11	15	15	1
OSPI (0–9)	11	11	11	0
PUF SI (0–20)	12	13	13	1
PUF PI (0–16)	6	5	5	0

She had no sexual intercourse. Therefore, the question of No. 3 is not included in the total score.

operation, she underwent the second surgery. The trigonal ulcer had recurred and electric fulguration was performed again. The symptom was relieved temporarily, similar to that after the first surgery. Weekly bladder injections of 50 mL of 50% dimethyl sulfoxide were continued for 10 weeks. However, her symptoms did not improve and she was unable to attend school. Thus, the third surgery was scheduled 7 months after the second procedure. During the third surgery, complete electric resection was performed for the recurrent trigonal ulcer (Fig. 3). After the third procedure, pain on urination disappeared (Table 1). She has had no pain without medication for 15 months postoperatively.

Discussion

Transurethral surgery, laser resection, and hydro-distention have been reported for the treatment of interstitial cystitis, especially for the ulcerative type.^{2,3} Electrical fulguration and resection are the two methods available for ulcer treatment. Peeker *et al.* retrospectively investigated the efficacy of transurethral ulcer resection in 103 patients, and observed improvement of symptoms in 92 patients.⁴ The treatment effects are considered to be due to the removal of nerve terminals that get activated during the inflammatory response.⁵ Based on this theory, electric fulguration was insufficient to remove the trigonal ulcer in our patient. Indeed, although two rounds of transurethral fulguration demonstrated no improvement, complete resection of the ulcer markedly reduced her pain. This suggests that total removal of the local lesion is important to stop the immune

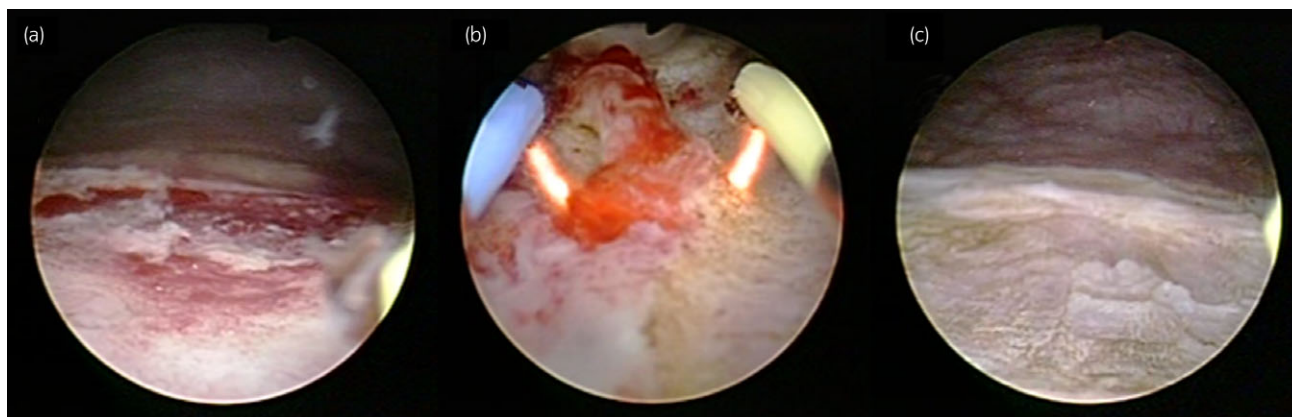


Fig. 3 Appearance of the trigone at the third operation. The trigonal ulcer recurred (a). Complete electric resection was carried out for recurrent trigonal ulcer (b). Appearance after complete resection (c).

reaction leading to interstitial cystitis. However, ulcers are often distributed widely, and it is difficult to identify the border between the ulcer and normal mucosa. Therefore, complete ulcer resection cannot be applied for all ulcerative interstitial cystitis patients. This point is the demerit of electrical resection. Kajiwara *et al.* reported the efficacy of narrow band imaging-assisted bladder surgery to precisely detect ulcerative lesions.⁶ In our patient, the border between the trigonal ulcer and normal mucosa was relatively clear, and narrow band imaging was not needed. Complete removal of the ulcer may be necessary for the treatment of ulcerative interstitial cystitis.

Conclusion

In conclusion, complete resection of the ulcer is effective for interstitial cystitis in adolescent female patients.

Conflict of interest

The authors declare no conflict of interest.

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