CASE REPORT

Coexistence of inflammatory pseudotumour and pseudomembranous cystitis: a histopathological surprise

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SUMMARY

We report a case of inflammatory pseudotumour (IP) and pseudomembranous cystitis (PC) in a young female patient who presented with gross haematuria and clot retention. Cystoscopy revealed a single glistening, polypoid growth of approximately 3 cm × 2.5 cm at the left lateral wall and the trigone of the urinary bladder. She was treated by transurethral resection of bladder mass. The patient's symptoms significantly improved after surgery. Histopathology report revealed haphazardly arranged myofibroblasts, fibroblasts and histiocytes and discrete areas of non-keratinising squamous metaplasia that was suggestive of IP coexistence with PC. She was doing well during the follow-up period of 12 months. Cystoscopy taken at 3 monthly intervals did not show any evidence of tumour recurrence.

BACKGROUND

A young woman who presented with intermittent gross haematuria and clot retention mimicking malignant tumour was diagnosed to have inflammatory pseudotumour (IP) and pseudomembranous cystitis (PC). PC is the term used to describe squamous metaplastic changes of the bladder trigone exclusively found in women. An IP of the urinary bladder is a benign lesion of obscure aetiology characterised by abnormal reparative response of fibroblastic spindle cells and granulation tissues to infection, trauma and surgery. We report the first case of IP coexistent with PC; however, the association between these two entities remains inconclusive.

CASE PRESENTATION

A young 20-year-old woman presented with a history of intermittent, painless gross haematuria associated with storage and obstructive lower urinary tract infections for 6 months. One day prior to admission, she went into clot retention for which clot evacuation and periurethral catheterisation was performed (outside). The symptoms aggravated cyclically prior to menstrual period. There was no history of fever, urinary tract infections, trauma or instrumentation.

INVESTIGATIONS

Physical examination was unremarkable. Initial investigations with renal and bladder ultrasound revealed single, smooth globular mass at the bladder neck approximately 3 × 2.5 cm in size with normal upper tracts (figure 1A). Renal function tests were within the normal limits. Urine routine examination showed 2–3 pus cells and 10–15 red blood cells, while urine culture was sterile.

Cystoscopy showed a single glistening polypoid growth involving the left lateral wall and trigone abutting the bladder neck. The contrast enhanced CT scan pelvis confirmed the presence of smooth contoured homogeneous enhancing mass 3 × 3 cm in size at the bladder neck (figure 1B).

TREATMENT

She underwent a complete transurethral resection of the bladder mass. The intraoperative and post-operative period was uneventful. Her symptoms were significantly improved after tumour removal. Histopathology showed haphazardly arranged myofibroblasts, fibroblasts and histiocytes intermingled with inflammatory cells. There were multiple patches of non-keratinising glycogenated squamous epithelium resembling vaginal epithelium. No malignant cells were seen (figure 2).

OUTCOME AND FOLLOW-UP

The patient was doing well after 1 year of follow-up with no evidence of recurrence on check cystoscopy that was conducted on three monthly intervals.

DISCUSSION

PC and IP are benign lesions that were first described by Heymann¹ and Roth,² respectively (1980). PC is although a common urological entity, found only among young women, is under-reported in the English literature due to its asymptomatic presentation. The exact aetiopathogenesis of PC is still unclear. The characteristic lesion is restricted to the trigone of the bladder possibly due to the oestrogenic effect. This could be explained by the fact that the trigone is anatomically and embryologically distinct from the remainder of the bladder, thereby allowing the trigonal epithelium to respond to oestrogenic stimulation.³ Pacchioni *et al*⁴ demonstrated a clear correlation between the presence of steroid receptors and squamous metaplasia of the trigone. Bacteria cystitis has been associated with the development of PC in cases of recurrent urinary tract infections.⁵

Similarly the exact aetiopathogenesis of IP is still obscure. It was supposed to be due to abnormal reparative response of fibroblastic spindle cells and granulation tissues to infection, trauma and surgery.⁶ IP is characterised histologically by the presence of acute and chronic inflammatory cells with a variable fibrous response. The radiological appearance of inflammatory pseudotumour is non-specific and

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Figure 1 (A and B) Ultrasound and contrast enhanced CT scan showing homogeneous enhancing mass at the bladder neck.

cannot be easily differentiated from a malignant neoplasm.^{7 8}

In the present case, gross haematuria with clot retention, enhancing lesion on contrast enhanced CT scan and cystoscopic appearance of the bladder growth were strongly pointing towards malignant tumour. But the final histopathology report was suggestive of IP coexistent with PC. The haematuria can be explained by the vascular nature and superficial ulceration of IP.⁹ Both the entities are indolent, possess no or low malignant potential and transurethral resection is almost curative, but a strict vigilance is mandatory as simultaneous occurrence of carcinoma in situ has been reported.¹⁰ This is the first case showing simultaneous existence of the PC and IP. Although no association between these two entities has been described in the literature, further studies are needed to demonstrate any possible association between IP and PC.

In conclusion, we report an index case of an IP of the urinary bladder along with PC presented with gross haematuria and clot retention. Clinical presentation and radiological studies were inconclusive, so the final diagnosis was solely based on histopathology. Also a long term follow-up with periodic cystoscopy should be considered. Every attempt should be made to distinguish between non-neoplastic proliferation and malignant tumour. Further studies are needed to prove any correlation between IP and PC.

Learning points

- ▶ Coexistence of inflammatory pseudotumour and pseudomembranous cystitis is a rare histological finding.
- ▶ Both entities are benign and possess no or low malignant potential.
- ▶ Accurate differentiation between non-neoplastic proliferation and malignant tumour of urinary bladder is mandatory.

Competing interests None.

Patient consent Obtained.

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Figure 2 (A and B) Histopathology showing scattered fibroblast, myofibroblasts and histiocytes along with areas of non-keratinising epithelium resembling vaginal epithelium.

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