CASE REPORT

Xanthoma of the urinary bladder: a rare benign condition which may be mistaken for malignancy

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SUMMARY

A 77-year-old man was referred with a history of recurrent urinary tract infections and a raised prostate-specific antigen. He was found to have an atypical lesion of the urinary bladder on cystoscopic examination. A preoperative MRI study suggested no evidence of malignancy and histology was consistent with urinary bladder xanthoma. This is a benign and exceedingly rare condition which requires no further treatment or follow-up. Patients should, however, have a lipid profile measured. Our case illustrates the use of MRI as an adjunct to aid operative planning. Although not essential, it may help to avoid an over aggressive initial resection in more precarious areas of the urinary bladder.

BACKGROUND

Xanthoma is a benign condition which appears clinically as yellowish lesions on the skin or the mucous membranes and is characterised histologically by aggregation of lipid-laden foamy macrophages. It occurs in the skin and tendons and is usually associated with hyperlipidaemia. Urinary bladder xanthoma (UBX) is rare with only 15 cases previously reported worldwide.^{1–9} The use of preoperative MRI suggested that the lesion was benign in nature. This imaging modality is not essential in evaluating abnormal bladder lesions. However, its use in this case allowed for a more conservative initial transurethral resection (TUR) to gain histological diagnosis, thereby reducing the risk of inadvertent resection of the ureteric orifice.

CASE PRESENTATION

A man in his mid-70s was referred to our outpatient urology department for an elevated prostate-specific antigen (PSA) of 6.7 ng/mL and a history of urinary tract infection. His medical history included atrial fibrillation, asthma, chronic kidney disease and TUR of the prostate in the 1980s. A digital rectal examination revealed a marginally firm prostate and a urine dipstick was positive for blood. A flexible cystoscopy showed an abnormal looking lesion on the left side of the bladder just beyond the ureteric orifice. Subsequently, the patient underwent a TUR of the lesion, presumed to represent an atypical form of bladder tumour. In view of a persistently raised PSA (9.6 ng/mL), a prostate biopsy was performed which showed histologically benign tissue.

INVESTIGATIONS

A preoperative MRI of the pelvis and the urinary bladder was performed. This study identified an area of focal thickening of the left bladder wall posterior and lateral to the insertion of the left ureter, with intermediate to low signal on the T2-weighted studies (figure 1). There was no focus of high signal within the bladder wall on diffusion-weighted imaging to suggest the presence of a malignant neoplasm. In conclusion, the MRI showed an area of minor focal thickening of the bladder wall of uncertain significance.

At TUR, a yellowish-white patch with multiple follicular projections was identified 1 cm lateral and posterior to the left ureteric orifice. In addition, a few smaller but macroscopically similar lesions were noticed over the left ureteric orifice (figure 2). With knowledge of the preoperative MRI, the patch was resected while the smaller lesions were left without intervention to avoid the risk of damage to the left ureteric orifice.

Histology showed papillary projections of submucosal tissue containing numerous foamy macrophages which stained positive for CD68 immunostains and was covered with unremarkable urothelium (figure 3). The histological and immunohistochemical features were consistent with UBX.

After the diagnosis was performed, blood tests revealed a normal fasting lipid profile.

OUTCOME AND FOLLOW-UP

The patient was reassured of the benign nature of UBX and further follow-up was arranged for his raised PSA but not for UBX.

DISCUSSION

The addition of this case to the total number of UBX reaches 16 worldwide, 9 in English, 5 in Japanese, 1 in Spanish and 1 in Korean.^{1–9} Lindboe² reviewed the first English report of UBX by Scholl in 1944 'Xanthoma and carcinoma in a diverticulum of the urinary bladder' and found that the histology was suggestive of xanthogranulomatous cystitis rather than UBX, due to the findings of multinuclear giant cells and infiltration by lymphocytes and plasma cells.¹⁰

Among all cases, there were 10 males and 6 females (1:0.6) with an average age of 62 years (range 47–80). UBX is almost always an incidental finding of cystoscopic examinations. UBX was seen in different sites of the bladder and described typically as a solitary yellowish-white plaque or patch

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Figure 1 MRI of the pelvis and the urinary bladder showing urinary bladder xanthoma (the arrow head) lateral and posterior to the insertion of the left ureter.

10–15 mm in diameter with an irregular or velvety surface. In five cases, a concomitant presence of, or a history of, low grade superficial transitional cell carcinoma was reported. Management varied and included eight TUR, two cystodiathermy, one partial cystectomy and five patients had only a cold cup biopsy.

UBX is a rare differential diagnosis of a white patch in the urinary bladder. Malakoplakia and xanthogranulomatous cystitis are both rare inflammatory conditions with similar microscopic features of UBX. Malakoplakia is characterised by large histiocytes (von Hanseman cells) and small basophilic inclusion structures (Michaelis-Gutmann bodies). Xanthogranulomatous cystitis is similar to malakoplakia but without Michaelis-Gutmann bodies. In UBX, the submucosa is packed with foamy macrophages in the absence of an inflammatory response. The use of CD68 as a macrophages marker in UBX was reported for the first time by Chitale *et al.*⁵

UBX can be added to the differential diagnosis of non-malignant bladder masses.¹¹ To the best of our knowledge,

we report the first radiological assessment of UBX. Although the MRI findings were non-specific, in our case it was accurate in excluding a malignant process. This was helpful in operative planning and facilitated the decision not to resect the lesion close to the left ureteric orifice lesion at the initial TUR, to avoid unnecessary risk of complications. The use of this imaging modality, however, should not preclude the need for histological diagnosis, but can act as a useful diagnostic tool.

Follow-up cystoscopy with no further action was reported in the previous literature and ranges between 5 and 12 months.^{3,4} UBX is a benign condition with no premalignant potential, and therefore follow-up with regular check cystoscopies is deemed unnecessary.^{4,5} Xanthoma can be a sign of underlying lipid metabolism disorder; however, only 3 of 16 cases of UBX reviewed were found to have hyperlipidaemia.¹ Nevertheless, patients diagnosed with UBX should have a lipid profile check.

Figure 2 Cystoscopic views of the bladder showing the patch of urinary bladder xanthoma (the double head thick arrow) lateral and posterior to the left ureteric orifice (the single head thin arrow) which is overlaid by yellowish white tiny spots.

Figure 3 Microscopic histology showing normal urothelium and the submucosal area contains numerous foamy macrophages (left, H&E $\times 200$) stained positive for CD68 immunostains (right, CD68 $\times 200$).

Learning points

- ▶ Urinary bladder xanthoma (UBX) is a rare incidental benign pathology with only 16 cases reported until now.
- ▶ It can be mistaken for malignant lesions during cystoscopy.
- ▶ MRI may be utilised to assess atypical lesions that occur in more precarious areas of the bladder to avoid over aggressive initial resection and reduce the risk of complications.
- ▶ Histology is the mainstay of diagnosis and transurethral resection or cold cup biopsy is essential.
- ▶ UBX is not a premalignant condition and long-term follow-up is unnecessary.

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