Novel treatment (new drug/intervention; established drug/procedure in new situation)

Phyllodes tumour of the prostate posing diagnostic and therapeutic dilemmas

Lucy Maudlin,¹ Richard Y Ball,² Stuart Irving,¹ Peter Preston,³ Sudhanshu Chitale¹¹ Department of Urology, Norfolk & Norwich University Hospitals NHS Foundation Trust, Norwich, Norfolk, UK² Department of Histopathology, Norfolk & Norwich University Hospitals NHS Foundation Trust, Norwich, Norfolk, UK³ Department of Radiology, Norfolk & Norwich University Hospitals NHS Foundation Trust, Norwich, Norfolk, UK

Correspondence to Sudhanshu Chitale, chitalenorwich@aol.com

Summary

A rare case of phyllodes tumour of the prostate of which less than 100 cases have been reported in the literature. It has an unclear clinical course and uncertain management options. In an inoperable case such as this, the authors discuss the difficulties faced with its management and the limited, possibly palliative role, of hormonal treatment using luteinising hormone-releasing hormone analogue in managing it over a medium term.

BACKGROUND

This is a rare pathology of prostate with unclear management options. In an inoperable situation, such as noted in our case, we describe difficulties faced at various stages of its management, the limited number of options available and the role played by a luteinising hormone-releasing hormone analogue (LHRH-a) in its overall management, which was largely palliative in nature.

CASE PRESENTATION

A Caucasian man in his mid sixties presented with acute urinary retention. His prostate specific antigen (PSA) level was markedly raised at 97 ng/ml ($N \leq 4.0$ ng/ml), and on clinical examination he had a grossly enlarged benign-feeling prostate gland. Transrectal ultrasound scan (TRUS) showed a prostate volume of 145 ml with multiple hypoechoic areas, indicating extensive cystic structures (figure 1A-B), but on biopsy there was no evidence of adenocarcinoma. No further investigations were arranged at this stage. A trial without catheter was successful and the patient's lower urinary tract symptoms (LUTSs) were well-controlled with α -blockers; hence, transurethral prostatectomy was not offered. PSA was monitored closely and the

patient was managed conservatively as he had multiple medical co-morbidities and had suffered a major cerebrovascular accident. Therefore, he was deemed unsuitable for aggressive/radical surgical treatment.

TRUS and prostate biopsy were repeated 3 years later as the PSA had gradually risen to 192.8 ng/ml. TRUS showed the same cystic findings. Histopathological findings, which resembled those seen on previous biopsy, revealed large atypical stromal cells, some multinucleate, with plentiful mitoses, including bizarre forms. The cells were immunoreactive for vimentin, but they did not stain for PSA, cytokeratins or CD68 (a macrophage marker). Several long, flat, surfaces lined by normal looking epithelial and basal cells were also present but there was no evidence of cancer although occult prostatic carcinoma remained a possibility. Based on the histological appearances, a diagnosis of phyllodes tumour (likely non-PSA secreting) was suggested (figure 2). A CT scan confirmed the TRUS findings and showed a prostate volume of about 1175 ml. Bone scan was negative for metastases.

The patient was considered for radical cysto-prostatectomy based on the evidence of a non-metastatic locally advanced extensive lesion, which warranted surgical

Figure 1 (A,B) Transrectal ultrasound scan showing extensive cystic changes in the prostate.

Figure 2 Histopathological section of prostatic tissue showing a bland epithelial surface with underlying bizarre stromal cells. H&E $\times 20$ objective.

Figure 3 (A,B) CT scan showing large prostatic mass with tumour necrosis/haematoma.

excision as the only means of disease control/cure. However, in view of the heightened risks associated with the procedure, he was deemed unfit for major extirpative surgery and as such, quite rightly, declined surgery.

Therefore, hormone treatment in the form of depot LHRH-a was started on account of his rising PSA to achieve androgen blockade.

The patient was re-admitted 4 months later with increasing LUTS and threatened urinary retention but neither a urethral nor a suprapubic catheter could be inserted because of the tumour's size. A CT scan showed a large (1300 ml) tumour filling the pelvis with the bladder lying anterior to the tumour and compressed between it and the anterior abdominal wall. There were areas of fluid, which could have represented cyst fluid, tumour necrosis or recent haematoma (figure 3A-B). Catheterisation of the bladder

failed because of the large tumour volume but the patient eventually managed to void spontaneously.

There was no ultrasound evidence of obstructive uropathy but the patient went on to develop acute-on-chronic renal failure as a result of cast nephropathy secondary to recently diagnosed plasma cell (multiple) myeloma. He was also found to have a 4×3 cm right hilar lung mass, which occluded the upper lobe bronchus leading to collapse of the lobe. It was associated with several enlarged lymph nodes in the carinal, precaval and paratracheal groups. The appearances were suggestive of advanced bronchogenic carcinoma but could have been metastases from the phyllodes tumour although no bronchoscopy and biopsy was performed to ascertain the exact pathology. Hypercalcaemia was noted presumably related to the plasma cell myeloma.

The patient was unfit for surgical excision of the prostatic tumour and palliative hormone treatment was continued as it had managed to lower the PSA down to 3.0 ng/ml (from 192.8 ng/ml) in 4 months, although it had failed to achieve reduction in the size of the prostate, thereby suggesting presence of a non-PSA secreting phyllodes tumour.

INVESTIGATIONS

PSA, TRUS scan, prostate biopsy, CT scan and bone scan.

DIFFERENTIAL DIAGNOSIS

In view of the typical features of phyllodes tumour of the prostate seen on the TRUS and CT scan, further confirmed on the prostate biopsy, we had no other differential diagnosis to consider except the possibility of co-existent prostate cancer, which remained unproven on two sets of biopsies.

TREATMENT

Androgen blockade treatment using depot LHRH-a.

OUTCOME AND FOLLOW-UP

Within 5 months of his recent admission—that is, altogether 9 months after starting hormonal treatment that had enabled him to reach a PSA nadir of 3 ng/ml—the patient died at home of chronic renal failure (with no evidence of obstructive uropathy) and complications of co-existent plasma cell myeloma.

DISCUSSION

Phyllodes tumour of the prostate gland is rare with fewer than 100 cases described worldwide.^{1 2} Management is uncertain and very few reported cases have been followed up long-term.³ The size of the tumour has varied from 3 g to 11.2 kg with variable PSA readings, which do not always reflect the tumour size. Phyllodes tumour of the prostate originates from the stroma and has uncertain malignant potential; although several case reports describe sarcomatous features or metastatic spread, there is as yet no single finding that reliably predicts clinical behaviour.^{4 5} It is unclear whether phyllodes tumours enhance the PSA uniformly or not. Immuno-histochemical studies show immuno-reactivity in epithelial cells for PSA.⁵ It is also reported that these tumours often express epidermal growth factor and androgen receptors; thereby, leading to

the suggestion that antiandrogen agents could well play a part in managing these patients.⁴

This case, which was followed up for up to 3 years and 9 months, clearly illustrates the difficulties faced in diagnosing and managing a rare prostatic tumour that had grown to become inoperable in a patient unfit for aggressive treatment. Most patients are managed with surgery and in those unsuitable for prostatectomy there is some evidence to suggest that chemotherapy can cause remission of phyllodes tumours.⁶

Antiandrogen hormone treatment may have a limited role if the tumour happens to be a non-PSA enhancing one but could be considered as a possible and, in the given circumstances, the most appropriate and perhaps the only feasible therapeutic option to achieve some regression of the background prostatic enlargement if not of the tumour itself unless it is a PSA-secreting tumour.

Learning points

- ▶ Phyllodes tumour of the prostate is rare with its diagnosis suspected on radiology but confirmed only on biopsy particularly when it presents with raised PSA.
- ▶ Treatment options and clinical course for phyllodes tumour still remain uncertain and unclear.

Competing interests None.

Patient consent Not obtained.

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