Sir,

Pseudoepitheliomatous, keratotic and micaceous balanitis (PKMB) is a rare, acquired disease of elderly men mainly involving the glans penis. It is characterized by progressive thick crusts and sheets of mica-like scales often leading to phimosis. It was first described by Lortat-Jacob and Civatte in 1961 (1). The exact aetiology is unknown.

The histological features include acanthosis, gross hyperkeratosis and hyperplasia with irregular downward extensions, with infiltration of chronic inflammatory cells (2–5). Once assumed to be a benign condition, there is now growing evidence that it should be viewed as a premalignant condition or a locally invasive low-grade malignant lesion (2–5).

We report a case of PKMB with the unusual involvement of the urethral meatus leading to urinary obstruction and physical disability as regards sexual function.

CASE REPORT

A 57-year-old Indian (Hindu) man presented with gradually progressive scaly eruptions on the glans penis for 1 year, which only caused mild local discomfort initially and progressed to cause difficulty in passing urine for the last 4 months. Of late, he noticed that there was splaying of the urinary stream like a fountain. Once in a while, he resorted to inserting a glass rod to obviate the obstruction of the urinary flow. He also found it difficult to perform the normal sexual act.

His past and family history did not reveal any significant illness. There was no antecedent history of long-term medications. He declared no history of extra-marital sexual exposure, genital ulcer, warts or urethral discharge for himself or his spouse. There was no history of trauma or instrumentation, but circumcision for unexplained phimosis had been done 8 years ago. He had no habits other than chewing tobacco for many years. There was no history of ocular symptoms, arthritis or cutaneous manifestations suggestive of Reiter’s disease. Therapy received earlier at other centres included courses of systemic antimicrobial agents (including penicillin, ciprofloxacin, cefotaxime, tinidazole, metronidazole), oral methotrexate, local and systemic corticosteroids, and surgical removal of the scales.

On examination, there were thick, dry, white and yellow mica-like sheets and keratotic masses adherent to the glans penis and the coronal sulcus. On removal of scaly plaques, a glazed erythematous surface was revealed. The urethral meatus had a hyperkeratotic yellow inner lining, as if a thick, tough membrane was plugged into the navicular fossa, which was thrown into the folds resulting in the formation of multiple sieve-like openings (Fig. 1). While voiding, three to four streams were seen.

There was no regional lymphadenopathy. There was no evidence of psoriasis, lichen planus or other related scaly dermatoses. Systemic examination was normal. Examination of the spouse did not reveal any evidence of genital wart or other sexually transmitted diseases.

The patient’s complete blood counts, ESR, blood glucose, liver and renal function tests and urinalysis were normal. HIV antibodies and venereal disease research laboratory tests were negative. Stool examination was normal. Elliptical biopsy from a thick keratotic lesion adjoining the coronal sulcus from the glans revealed acanthosis, massive hyperkeratosis, pseudoepitheliomatous hyperplasia and a sparse mononuclear cell infiltrate but with no frank malignancy or cellular atypia. Photodocumentation of the histopathology was not available.

He was treated with topical 5-fluorouracil cream 5% for 6 weeks. Although the heaps of scales were ablated to some extent, the local discomfort, urinary obstruction and difficulty in sexual activities showed no symptomatic remission. Urethral dilatation with metal bougies and, subsequently, a topical preparation containing clobetasol propionate and salicylic acid did not help much in ameliorating his urethral symptoms. We could not try lasers or radiation therapy, as these are unavailable at our centre. The patient declined our proposal for further surgical intervention, and was subsequently lost for follow-up.

DISCUSSION

PKMB is an uncommon disease. The major concern is its association with verrucous carcinoma of the penis (5, 6). However, there was no evidence of malignancy in...
our patient. Repeat biopsy is justified at a later date to detect a possible malignant change.

The association of PKMB with circumcision is well known. Our patient developed PKMB 7 years after the circumcision. In most cases of uncircumcised elderly males, phimosis supervenes (1) and circumcision is then indicated, whereas in many cases PKMB developed after circumcision carried out late in life (2). Therefore, whether circumcision is the cause or the effect of PKMB remains to be elucidated.

His symptoms of urinary obstruction developed later in the course of the disease as the urethral meatus became involved. Progressive hyperkeratosis of the mucosal lining led to compartmentalization of the urethral opening into multiple slits. This resulted in urinary flow as multiple streams. To the best of our knowledge, such a complication has not been reported for PKMB. We propose the term ‘watering-can penis’ to describe such a presentation.

Multiple streams of urine and the feeling of obstruction led to great psychological distress and embarrassment, particularly when voiding at public restrooms. We believe that his difficulties in sexual intercourse are related to a loss of elasticity at the glans and coronal sulcus due to severe hyperkeratosis and narrowing of the external meatus.

Factors leading to such an unusually severe urethral involvement in this case remain unknown. We believe that the underlying pathology is the same as that affecting the adjoining genital mucosa, as there was no evidence of any other concomitant disease.

Tobacco chewing, like smoking, is a significant risk factor in the carcinogenesis of penile carcinoma in India. N-Nitrosamines, polycyclic aromated hydrocarbons and polonium-210 are recognized potential carcinogens. The cumulative effect of these factors, either directly or their metabolites in the smegma, reduction of immune surveillance by reducing Langerhans’ cell number and induction of mutagenicity by nitrosamines are some of the mechanisms implicated (6).

PKMB has been described to go through four clinical stages – plaque stage, verrucous tumour stage, transformation into verrucous carcinoma, and invasive stage with possible lymph node involvement and metastasis. It has been suggested that the plaque stage can be managed by topical 5-fluorouracil. Other stages should be managed by local surgical excision or partial penectomy with more radical surgery depending on the extent of involvement. Other modalities of treatment include cryotherapy, X-ray irradiation, shave biopsy plus electrocoagulation and CO₂ laser therapy (7).

We believe that our patient was still in the initial plaque stage with unusually severe urethral involvement. However, we were not successful in significantly relieving his urethral symptoms by the various non-invasive therapies attempted. We did not favour electrocoagulation or cryotherapy for the urethral lesions for fear of further worsening of obstruction of the urethral meatus.

We believe that the further therapeutic option in this case would have been surgical excision with reconstruction of the external urethral meatus. We hoped that such a procedure might render at least symptomatic remission and functional improvement. However, recurrence of PKMB lesions 1 year after excision has been reported (8).

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