Scrotal elephantiasis secondary to recalcitrant hidradenitis suppurativa

Sir,

Hidradenitis suppurativa is a complex and challenging disease, the severity of which ranges from almost asymptomatic to one that severely affects the quality of life.^[1] Clinicians often face severely affected patients who require a multi-disciplinary approach for diagnosis and treatment. We report a patient not responding to conventional therapy, who was then treated with the help of a plastic surgeon experienced in genital anatomy.

The patient was a 40-year-old Caucasian man who had hidradenitis suppurativa since the age of eighteen. The diagnosis was made on the basis of clinical features (typical lesions, abscesses, sinus, scars, nodules, typical topography, chronicity and recurrence). The axillary involvement had always been mild with scarring seen at the time of presentation. However, the perineal region was severely affected with scrotal elephantiasis and chronic suppuration [Figure 1a and b] despite various medical and surgical treatments during the course of disease. He was treated with various antibiotics, systemic retinoids, etanercept and also underwent incision and drainage of the lesions. The patient was married and sexually active but had no children. He had never visited countries endemic for filariasis.

He was admitted to our ward with significant scrotal swelling, pain and suppuration which hampered his ability to walk. He was treated with metronidazole and moxifloxacin for several weeks prior to admission but his symptoms continued to worsen. Physical examination also revealed condylomata acuminata over the penis, perineum and inguinal areas which we treated with carbon dioxide laser.

Complete blood chemistry revealed microcytic, hypochromic anemia with decreased hemoglobin (8.2 gm/dl), decreased serum iron and normal ferritin levels. Screening for human immunodeficiency virus was negative. A magnetic resonsnance imaging (MRI) scan revealed exuberant lymphedema of the scrotum, inguinal area and perineum with numerous sinus tracts [Figure 1c]. Microbiological culture of the exudate revealed polymicrobial flora with no evidence of fungus or mycobacteria. Colonoscopy was done and biopsies were taken from the colon and ileum to rule out inflammatory bowel disease.^[2]

The patient was treated with intravenous imipenem/ cilastatin 1 gm thrice daily. There was reasonable improvement of scrotal suppuration and pain. The modified hidradenitis suppurativa score (Sartorius and Lapin) was 129. Since the patient was unsatisfactorily treated with etanercept earlier, other TNF alpha inhibitors such as infliximab or adalimumab^[3] were not considered. We considered a surgical approach with the help of a plastic surgeon and urologist. Complete excision of the affected tissue beyond the borders of activity was performed, leaving clear margins.^[4] The surgical defect was then covered with partial-thickness skin grafts from the thighs [Figure 1d]. The extensive scrotal fibrosis rendered testicular identification impossible, even with intraoperative ultrasonography. Pathological examination of the excised piece did not find any testicular tissue either. Testosterone levels were repeated several times during follow up over a period of one year, and were always within normal limits. There was no change in secondary male characteristics. libido and erectile function.

We believe that acceptable results with conservative therapeutic options were not possible in this case. Scrotal elephantiasis secondary to hidradenitis suppurativa is rare and highly disfiguring.^[2,3,5] In our patient, it followed an insidious and progressive course in spite of various pharmacological therapies. Severe

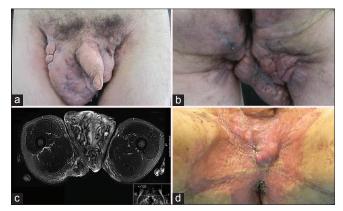


Figure 1: (a) Scrotal and penile lymphedema with multiple suppurative sinus tracts (b) Lesions in the gluteal and perineal region (c) MRI scan showing numerous sinus tracts and scrotal diameter enlargement (d) Postoperative appearance after four months showing no evidence of suppuration with an excellent functional and acceptable aesthetic outcome

hidradenitis suppurativa has a profound negative functional and aesthetic impact in patients. Surgical intervention improved the quality of life of this young patient, both socially and professionally.

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