Framboesiform lesions in primary herpes simplex infection:

A case report

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SUMMARY A 27 year old homosexual man developed unusual sacral lesions during a disseminated primary herpetic attack, which was confirmed by viral culture and rising antibody titre. The lesions had a striking framboesiform appearance and healed without ulceration or scarring. Review of modern and historical published reports suggests that this may be the first illustrated description of such infection.

Introduction

Disseminated herpes simplex infection usually occurs in the form of a severe systemic illness in atopic or immunocompromised people, who often die.^{1 2} This report concerns a case of multiple cutaneous lesions of framboesiform appearance in a patient who remained clinically well.

Case report

A 27 year old homosexual male musician presented with a three day history of penile and sacral blistering, slight dysuria, and groin pain eight days after active and passive oro-ano-genital intercourse with a casual partner who had an ulcerated oral lesion. Four weeks previously he had ended a two year monogamous homosexual relationship. He had had no prior oral or genital herpetic infection, or other sexually transmissible disease (STD). Apart from longstanding acne vulgaris treated with oxytetracycline 250 mg twice daily, there was no relevant medical history, drug treatment or abuse, and no systemic complaint.

On examination the patient appeared to be well and was not feverish. There was a single herpetiform ulcer on the right upper lip, and two umbilicated vesicles 8 mm in diameter surrounded by a slightly erythematous flare on the glans penis, with ten identical lesions scattered over the buttocks.

Bilateral, exquisitely tender inguinal lymphadenopathy was the only other abnormal finding. After routine screening for STD (see below) a provisional diagnosis of primary genital herpes was made and the patient was given idoxuridine 0.5% ointment and advised to bathe with physiological saline, to abstain from sexual intercourse, and to return for review.

Three days later he remained generally well, despite a severe deterioration in his skin condition. The penile blisters had ulcerated, leaving a painful meatitis and urethritis. Each sacral lesion had developed into an irregular, vividly erythematous raised plaque covered with clusters of vesicles with a striking framboesiform appearance (fig 1). The lip lesion had crusted over, there was ulceration of the right anterior border of the tongue and contiguous buccal mucosa, and a severe recrudescence of acne vulgaris, with a further framboesiform cluster on the neck (fig 2). General and central nervous system examination, with particular attention to trigeminal and sacral motor and sensory function, was entirely normal.

On review seven days after his initial presentation, the penile ulcers were crusting over, and the clustered vesicles in the sacral lesions had discharged without ulceration leaving a raised, dry, erythematous plaque that caused mild pruritus. The urethritis and meatitis were improving, and the inguinal lymph nodes were less prominent. After a further seven days most lesions had completely resolved, with a few lightly adherent crusts and no residual scarring. The acne was quiescent and there was no lymphadenopathy or dysuria. The patient had remained well throughout, having complained only of embarrassment about his facial appearance.

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FIG 1 Close up of lesions showing raised erythematous base, fluid filled vesicles (transparent), discharged vesicles (opaque), and brown crusts at initial site of blistering.

FIG 2 Framboesiform lesion on neck with facial acne vulgaris.

Investigations

Herpes simplex virus (HSV) type 1 was cultured from the lip, tongue, neck, sacrum, glans, and urethra on Vero cells, and the cultures were observed for 21 days to exclude infection by varicella zoster virus (VZV). The HSV complement fixing antibody titre rose from less than 1/10 to 1/40 over two weeks. The haemoglobin concentration was 14.5 g/dl, the white cell count was $17.3 \times 10^9/l$ with a polymorphonuclear leucocytosis, and the erythrocyte sedimentation rate was 8 mm in the first hour. Swabs transported in Stuart's medium grew normal skin flora only. Urine analysis was normal. At the first two visits the Gram stained urethral smear showed polymorphonuclear leucocytes, but urethral, rectal, and pharyngeal cultures gave negative results for *Neisseria gonorrhoeae*. The Venereal Disease Research Laboratory (VDRL) and *Treponema pallidum* haemagglutination (TPHA) serological tests, tests for hepatitis B surface antigen and hepatitis Be antigen, and repeated dark ground microscopy for treponemes, all gave negative results.

Discussion

The umbilicated vesicles seen on presentation appeared in retrospect to be identical with the varicelliform eruptions first described by Martin³ and Kaposi,⁴ and originally named "acute vesicular eczema" by Unna.⁵ Later the widespread distribution of these lesions suggested disseminated herpes simplex infection, which occurs principally in atopic people¹ and in those who are immunocompromised.^{2,6} With rare exceptions,⁷ this is accompanied by severe systemic symptoms causing high mortality, as in the cases originally presented by Hebra.⁸ The appearance of these lesions closely matches the original classification and description of the framboesiae by Sauvages de la Croix in 1763⁹: "Hi vero fungi sive minores, sive majores sunt coloris rosei, vel pallide rubri, granulosi, seu papillis exasperati, muco rufescente continuo madidi, nulli ulceri sed cuti adhaerentes."

The sharply defined erythematous base and clustered vesicles seen in the plaques are common features of infection, with VZV, which has been accurately described in many historical texts,^{4,5,10,11} and excellently illustrated by the lithographs of Bateman produced in 1817,¹² and by those of the Sydenham Society.¹³ Framboesiform lesions occur classically in the tropical treponematoses,^{4,9,11} and reminders that they may also occur in secondary syphilis have recently been published.^{14,15}

This patient had no personal or familial history of atopy, no history, signs, or symptoms suggestive of the acquired immune deficiency syndrome (AIDS), and no serological evidence of treponemal disease. The simultaneous appearance of the lesions at multiple scattered sites of inoculation, with a greater than fourfold rise in HSV antibody titre, indicates primary infection. We considered using acyclovir¹⁶ but, as the nervous system was not affected, there was no systemic illness, and the lesions resolved rapidly, this agent appeared to be unnecessary.

We have been unable to discover any previous report of this condition, and therefore conclude that this may be the first illustrated description of framboesiform lesions due to multiple primary mucocutaneous inoculation of herpes simplex virus type 1.

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