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CASE REPORT

Rapidly growing orf in a renal transplant recipient: favourable outcome with reduction of immunosuppression and imiquimod

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Summary

Orf is a viral skin infection due to a poxvirus. It manifests as a nodule of the hands that heals spontaneously within 3–4 weeks, but may be persisting and difficult to treat in immunocompromised patients. Very few cases have been reported in transplant patients; therefore, management is not established. We report a renal transplant recipient with a rapidly growing orf which regressed after application of imiquimod and a reduction in immunosuppression without damage on his renal function. This case suggests that a rapidly growing orf in transplant patients behaves as an opportunistic infection and therefore minimization should be considered along with a topical treatment.

Introduction

Orf, also known as ecthyma contagiosum, is a viral skin infection caused by a poxvirus. The disease is endemic in sheep and goats, but affects humans occasionally. It manifests as a cutaneous nodule appearing most commonly on the hands. It usually regresses spontaneously within 3–4 weeks, but may be persisting, extensive and difficult to treat in immunocompromised patients [1–3]. Very few cases of orf have been reported in transplant patients; therefore, the management of this uncommon infection is not yet established.

Case report

A 61-year-old man of North African origin received a kidney transplant in July 2005. After an acute rejection, which was successfully treated with a corticosteroid pulse in October 2005, his renal function remained stable and the patient was treated with prednisolone (10 mg/day), mycophenolate mofetil (2 g/day) and cyclosporine (150 mg/day). He was referred to us in February 2006 for a verrucous nodule surrounded by a purple border on the right forefinger, which had appeared 2 weeks earlier (Fig. 1a). The patient had been in contact with a freshly slaughtered sheep on the occasion of the celebration of Aid-el-Kebir 1 week before the onset of the lesion. The diagnosis of orf was made clinically and because of the recent and indolent character of the lesion, no treatment was given. Three weeks later, the patient consulted again because of the rapid growth of the nodule, which involved the whole median phalanx and had acquired a tumoural aspect, reminiscent of a giant pyogenic granuloma (Fig. 1b). Histological examination revealed necrosis of the upper epidermis with ulceration and eosinophilic cytoplasmic inclusions in some vacuolated epidermal keratinocytes. The dermis contained areas of oedematous granulation tissue and abundant thin-walled blood vessels (Fig. 1c). These findings were diagnostic of orf containing pyogenic granuloma-like changes. The lesion continued to grow

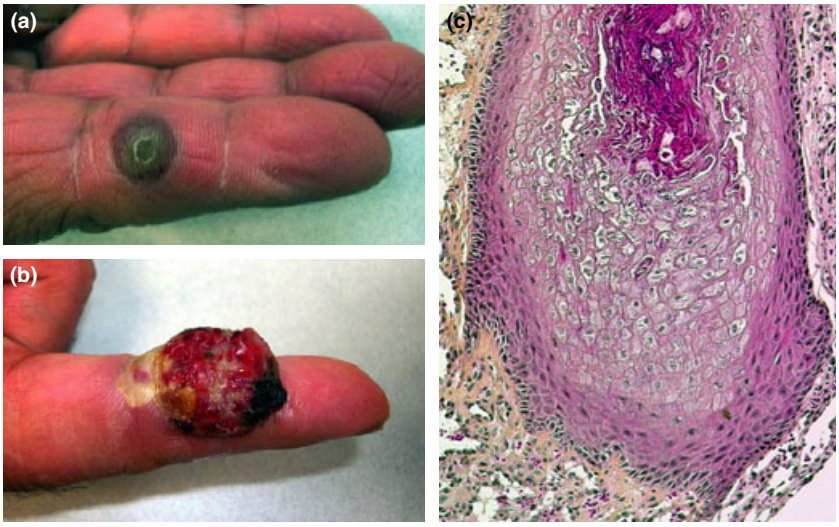

Figure 1 (a) Initial presentation of a typical orf. (b) Tumoral course of giant pyogenic granuloma-like orf. (c) Typical histological aspect of orf showing eosinophilic cytoplasmic inclusions in some vacuolated epidermal keratinocytes [lesion shown in (b)].

rapidly within the following week. Concomitantly, extensive dermatophytosis of the legs and toes developed. These manifestations suggested deep immunosuppression; therefore, the immunosuppressive treatment was decreased (prednisolone from 10 to 7.5 mg/day, and mycophenolate mofetil from 2 to 1.5 g/day) and local treatment of orf with imiquimod 5% cream was started. The medication was applied in escalating doses to avoid local intolerance, (every other day during the first week, daily during the following 2 weeks and twice daily for the next 3 weeks). The treatment was discontinued after 6 weeks because of substantial inflammation and decrease of the lesion. Complete healing was obtained 6 weeks later. Dermatophytosis was successfully treated with topical terbinafine. The patient had no further orf recurrences; his renal function has remained stable over the following 4 years; immunosuppression was not increased back to the initial doses.

Discussion

Orf affects either occupational groups such as animal breeders, veterinarians, or Muslim patients on the occasion of ritual sacrifice. Although organ transplant recipients are at high risk of viral disorders, orf has been seldom reported in this patient group [4–8]. In all cases, lesions were extensive, did not regress spontaneously and were always difficult to treat because of their location on the fingers. Treatments included repetitive surgery or cryotherapy because of iterative recurrences, or exceptional costly treatments such as cidofovir cream [6]. All patients were under triple treatment combining corticosteroids, calcineurin inhibitors and azathioprine or mycophenolate mofetil, but minimization was not considered.

Our case illustrates the extensive course of orf in a transplant patient, which was controlled both by a moderate reduction in immunosuppression and by applications of imiquimod. This immune-response modifier was used because of its antiviral activity, shown by good results achieved on molluscum contagiosum infection, which is also caused by a poxvirus. As the two measures were concomitant to hamper the rapid growth of the lesion, it is difficult to know which one played the predominant role in the regression of the lesion. Recently, imiquimod was reported successful for the treatment of recurrent orf in a renal transplant recipient, but in that case, the medication was used as an adjuvant therapy after surgical excision [8]. That lesion had developed some days before kidney transplantation and its growth had been favoured by the loading doses of immunosuppressants at grafting. Whether orf can regress spontaneously in transplant patients remains unknown as it seems likely that only severe infections are reported, probably reflecting deep immunosuppression as in our patient, who did not develop rejection after minimization. Indeed, the course of the most common viral infections (such as herpes simplex, herpes zoster or warts) may be similar to the nonimmunosuppressed population in transplant patients with stable immunosuppression.

The favourable outcome in our patient makes us believe that transplant patients developing orf should be closely monitored and, in case of rapid growth, minimization should be considered, in association with application of imiquimod. The current observation could potentially change practice patterns by avoiding surgery.

Authorship

DZ, CP-N, SE: followed the patient. JK performed the pathological study. DZ, JK, CP-N, SE: wrote collectively the paper.

Conflict of interest

Sylvie Euvrard received a travel grant from Meda Pharma for a congress and Jean Kanitakis is member of the advisory board of Meda Pharma.

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