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Extensive orf infection in a toddler with associated id reaction

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Abstract

Orf is a zoonotic parapoxvirus typically transmitted to humans by a bite from goats or sheep. We present an unusual case of multiple orf lesions on the fingers of a 13-month-old child who was bitten by a goat and subsequently developed progressive swelling, blistering, and necrotic papulonodules of the hand followed by an additional diffuse, pruritic, papular rash. A primary diagnosis of orf infection was confirmed using real-time polymerase chain reaction, and the diffuse eruption was clinically consistent with an id reaction. Extensive necrosis and papular id reaction associated with orf rarely have been described.

Keywords

child; contagious pustular dermatitis; ecthyma contagiosum; goat; infection; orf; parapoxvirus; virus

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1 | INTRODUCTION

Orf virus, also known as contagious ecthyma, contagious pustular dermatitis, sore mouth disease, and scabby mouth disease, is a zoonotic virus primarily affecting sheep and goats. Humans may be infected by direct contact with affected animals or fomites. The infection is most often reported in veterinarians, butchers, and farmers.[1–3] Orf infections in children are less common but have been reported.[2,4] Typically, affected individuals develop a single, circumscribed lesion on the hand that has contacted infected material.[1] We present an unusual case of a 13-month-old boy who developed multiple necrotic orf lesions and a generalized rash after a goat bite.

2 | CASE REPORT

A 13-month-old boy presented to the emergency department with multiple, raised, weeping blisters on the dorsum of his left hand. Two weeks before, a baby goat kept in a pen at his family's home had bitten four fingers of the boy's left hand.

The bite produced six open abrasions that became swollen and erythematous during the first few days after the bite and progressed into blistering at the site of the abrasions. One week after the bite, the child developed a low-grade fever and was evaluated by his pediatrician, who cultured the wound and prescribed a 7-day course of amoxicillin/clavulanic acid because of concern for bacterial infection, although the bacterial cultures were ultimately negative. When the lesions continued to worsen despite antibiotics, the child received an intramuscular injection of cefazolin and was sent to the Rady Children's Hospital-San Diego emergency department. Physical examination revealed six raised, blisterlike papulonodules with overlying serous crust and necrotic tissue on the dorsum of the fingers of the left hand. The lesions were minimally tender to palpation. The child was using his hand normally for grasping and displayed no guarding of the hand. He was afebrile and otherwise systemically well, with normal energy and appetite. With characteristic lesions on the fingers 2 weeks after the bite of a goat in an otherwise-healthy child, orf was considered a possible etiology.

The boy was admitted for further evaluation, including blood and wound cultures. Complete blood count was notable for leukocytosis with high absolute neutrophil count and erythrocyte sedimentation rate. Intravenous clindamycin and ceftriaxone were administered. An x-ray performed because of concern for osteomyelitis showed significant soft tissue swelling but no osseous abnormality. Samples of superficial crust and blister fluid from the affected hand were sent to the Centers for Disease Control and Prevention (CDC) for real-time polymerase chain reaction (PCR) diagnostic testing. Blood cultures remained sterile and wound cultures were positive for normal skin flora. The child was sent home on a 10-day course of clindamycin because of concern for secondary bacterial infection of his open wounds.

The following day the child developed a new diffuse itchy rash and a low-grade fever of 100.4°F and returned to the emergency department, at which time dermatology was consulted for further evaluation. According to the history, the primary lesions on the boy's dorsal left hand were unchanged from discharge the day before (Figures 1 and 2). The new

diffuse pruritic eruption consisted of multiple monomorphic, pink, 1- to 2-mm papules concentrated on bilateral hands and forearms (Figures 1, 2, and 3A), but also present on the chest, abdomen, back (Figure 3B), and legs (Figure 3C). The child appeared otherwise well. Based on the morphology of the lesions and timing of the eruption, the patient was diagnosed with an id reaction to the orf virus. Clindamycin, which had been started 2 days before the new rash, was also discontinued, because of the possibility of a drug reaction, and replaced with cephalexin. Triamcinolone 0.1% ointment and diphenhydramine were also prescribed for symptomatic treatment of the pruritus associated with the presumed id reaction.

On follow-up in the dermatology clinic 1 week later, the primary lesions had improved, with reduced swelling, erythema, and crust (Figure 4). The diffuse eruption was also resolving, with pinpoint crusted papules (Figure 5) that were no longer pruritic. The real-time PCR results from the CDC confirmed orf virus as the etiology of the primary papulovesicular lesions.

3 | DISCUSSION

Orf is a deoxyribonucleic acid parapoxvirus that primarily affects goats and sheep as a papulovesicular eruption on the mouth, nose, and limbs but can be transmitted to humans.[2] Although it is most often transmitted to humans by occupational exposure, children may be exposed to the disease on family farms or in recreational settings.[2]

Orf infection typically presents as a single, circumscribed 2- to 3-cm lesion that progresses through six clinical stages, each lasting roughly 1 week: (i) maculopapular stage, in which a red macule develops and then becomes elevated; (ii) targetoid stage, in which the red center of the lesion is surrounded by a white ring and an outer red halo; (iii) acute stage, in which a weepy, red nodule develops; (iv) regenerative stage, in which the lesion dries out and black dots composed of pyknotic cells form on its surface; (v) papillomatous stage, in which dry, verrucous papillations may develop on the surface of the lesion, resembling an exuberant wart; and (vi) regressive stage, in which the lesion flattens and a dry crust forms and eventually falls off.[1,5] Lesions most often occur on the hands[1] but have also been described elsewhere, including the face[1,2] and perianal area.[6] Multiple lesions are uncommon in immunocompetent individuals[1] but have been reported[1,2] and infrequently may be accompanied by malaise, fever, lymphangitis, and lymphadenitis.[1] Dermoscopy of orf lesions reveals well-defined nodules with central crust, stucturelessness white areas, and shiny white streaks. Lesions are surrounded by dotted and hairpin vessels and have fine peripheral scale.[7]

Skin biopsy of an orf lesion demonstrates varying histologic features depending on the stage of infection. The maculopapular and target stages show vacuolated epidermal cells with eosinophilic intracytoplasmic inclusions. During the acute stage, the epidermis degenerates, and multilocular vesicles are seen. During the regenerative stage, extrusion of pyknotic cells and regeneration of the epidermis are visualized. The papillomatous stage shows acanthosis with fingerlike downward projections of the epidermis through the full thickness of the dermis. In the regenerative stage, acanthosis diminishes and collagen is regenerated.[1,5]

Laboratory testing is not routinely performed in cases of suspected orf, but in several case series, complete blood counts were normal in all cases.[8,9]

The differential diagnosis for orf infection includes cutaneous anthrax,[5,10] which is also transmitted from goats but is typically more hemorrhagic, with more significant necrosis and eschar formation.[5] Milker's nodule (pseudocowpox), a parapoxvirus transmitted by cows, and cowpox, an orthopoxvirus transmitted primarily by small rodents, are clinically and histologically indistinguishable from orf.[5] Orf lesions may resemble herpetic whitlow but are typically less painful.[5] Sporotrichosis and tularemia are also in the differential diagnosis, but typically are accompanied by ascending lymphangitis that is uncommon in orf.[2,5] The absence of lymphadenopathy also helps distinguish orf from *Mycobacterium marmum* infection and leishmaniasis.[2] Syphilitic chancre, pyogenic granuloma, keratoacanthoma, giant molluscum, and primary inoculation of tuberculosis should also be considered.[5,7]

Usually orf virus is diagnosed clinically based on appearance and a history of contact with goats. Definitive confirmation of orf virus infection can be made using real-time PCR, available through the CDC Poxvirus Laboratory (https://www.cdc.gov/laboratory/specimen-submission/detail.html?CDCTestCode=CDC-10383). Turnaround is 1-5 business days after specimen receipt.

Orf infections typically resolve spontaneously in 4-6 weeks with minimal scarring.[1] Accordingly, therapy is primarily symptomatic.[3] Immunocompromised patients may be unable to clear the viral infection and may require treatment with topical cidofovir[11] or imiquimod.[12] Lesions also have been treated with shave excision, curettage, cryotherapy, and electrocautery.[3]

Complications of orf infection include secondary bacterial infection,[13] erythema multiforme, which develops in 7%-18% of cases,[14] vesiculopapular or papular eruptions,[13,15] and bullous pemphigoid-like eruptions.[13] Rare complications including giant orf lesions[3] and Stevens-Johnson syndrome have also been reported.[8]

The monomorphic, pink, papular morphology of the secondary eruption in this case was most consistent with an id reaction to the primary infection. An id reaction, also known as autoeczematization or disseminated secondary eczema, is a typically symmetric eruption of eczematous lesions distant from the site of a primary dermatosis. It is most often associated with allergic contact dermatitis and stasis dermatitis but also occurs in conjunction with fungal, molluscum, scabies, and other infections. Occasional development of a generalized, nonspecific allergic papular rash on the hands and feet in orf was described in a 1983 summary of the epidemiology of orf in New Zealand,[15] although to our knowledge, no specific cases of monomorphic papular id reaction to orf have been described in the literature. Nonetheless, given that id reaction is associated with molluscum, a member of the poxvirus family, its association with the orf parapoxvirus is perhaps unsurprising.

Limitations of this investigation include the lack of confirmation of orf infection in the goat and the inability to evaluate whether the goat was coinfected with other pathogens. Additionally, the presumed id reaction was not confirmed histologically.

4 | CONCLUSION

Orf is a parapoxvirus that typically infects ungulates and may be transmitted to humans through direct contact with animals or fomites. Orf infection occurs most commonly in adults through occupational exposure but has also been reported in children. Orf should be a primary consideration in the setting of a goat bite. This case was unusual because of the young age of the patient, multiple orf lesions on several fingers, significant eschar, and the associated monomorphic papular id reaction.

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FIGURE 1.
Erythematous, crusted papulonodules on the dorsal fingers. A diffuse eruption of 1- to 2-mm papules is also present

FIGURE 2.
Side view of the papulonodules highlighting their thick crust

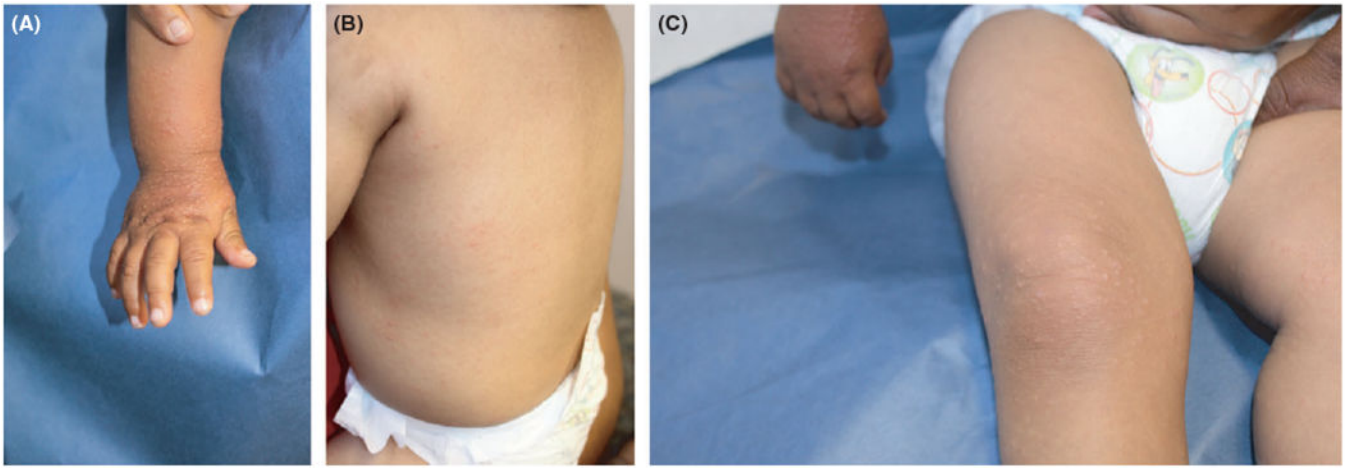

FIGURE 3.

Diffuse, papular, pruritic eruption that developed 11 days after the primary orf lesions. (A) The secondary papular eruption is most concentrated on the hands and forearms but is also present on the (B) back and (C) legs

FIGURE 4.
Primary orf lesions with reduced swelling, erythema, and crust 3 weeks after a goat bite

FIGURE 5.
One week after development, the diffuse secondary eruption has flattened into a pinpoint crust and is no longer pruritic