A child with lumps and bumps down on the farm

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CASE PRESENTATION

A 21-month-old Caucasian female with a history of eczema was referred to the pediatric surgery clinic for evaluation of bilateral axillary and right inguinal lesions of four months' duration. The lesions varied from 0.5 cm to 1 cm in diameter and were red in colour, fluctuant and nontender (Figures 1 and 2). There had been no response to oral antibiotics, and the left axillary lesion had been spontaneously draining pus (Figure 2). The child remained well and active, with no history of fever, night sweats, weight loss or cough. She had sustained a hamster bite to one of her fingers a few weeks before the appearance of these lesions. Her family operated a potato farm that had dogs and cats but no large farm animals. There was no history of travel or exposure to tuberculosis. The remainder of the physical examination was unremarkable, and her complete blood cell count and serum immunoglobulin levels were normal.

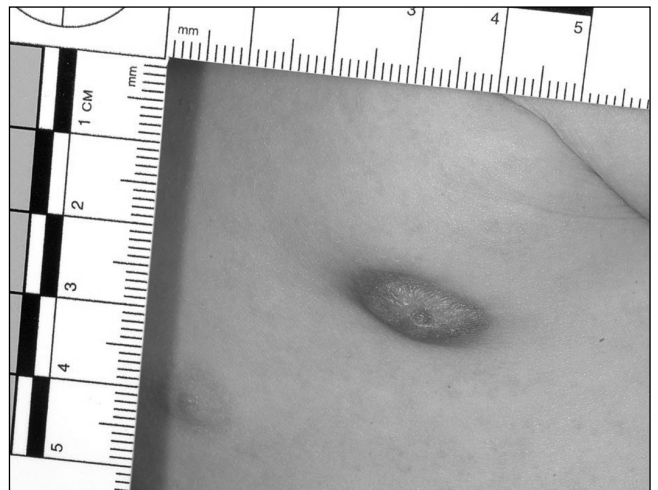What is the diagnosis?

Figure 1) *Right axillary lymph node*

Figure 2) *Left axillary lymph node with spontaneous drainage*

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DIAGNOSIS

The diagnosis was nontuberculous mycobacterial (NTM) lymphadenitis.

A tuberculin skin test (purified protein derivative) was positive at 20 mm. Based on this test result, the appearance of the lesions and the absence of a history compatible with exposure to tuberculosis, a presumptive diagnosis of NTM infection was made. Because there was no significant resolution of the lesions after a one-month course of azithromycin, surgical excision of all three lesions was performed. Histopathology showed granulomatous inflammation and extensive caseation. *Mycobacterium avium* complex (MAC) was first detected after nine days of incubation, identified by DNA probe and, ultimately, culture. The child developed presumptive secondary bacterial infection of one of the axillary sites. This rapidly resolved with antibiotics, and she remained well eight months later, with initial marked improvement in her eczema.

DISCUSSION

Lymphadenitis is the most common manifestation of NTM infection in children, generally occurring in otherwise healthy children younger than five years of age. MAC is most commonly linked to this condition, although *Mycobacterium scrofulaceum* was the predominant etiological agent when the disease was first recognized (1). Other mycobacterial species isolated from patients with lymphadenitis include *Mycobacterium fortuitum*, *Mycobacterium chelonae*, *Mycobacterium malmoense*, *Mycobacterium kansasii*, *Mycobacterium intracellulare*, *Mycobacterium xenopi* and *Mycobacterium haemophilum* (2). Because the specific culture methods required for the recovery of M *haemophilum* are not routinely employed, it is only since the advent of nucleic acid amplification testing methods that it has become evident that M *haemophilum* may be the next most common species, after MAC, to cause lymphadenitis (2). Future studies employing nucleic acid amplification testing may show geographic variability in the predominant species causing NTM lymphadenitis, as exposure to swimming water appears to increase the risk of M *haemophilum* infection (3).

NTM organisms are ubiquitous and can be isolated from soil, water, food products, plant material, birds, and domestic and wild animals (1). Tap water is considered to be the major reservoir for most common human NTM pathogens, and, as such, is of increasing public health interest (4). Transmission occurs through inhalation, ingestion or direct contact with environmental sources, and it seems unlikely that the hamster bite played any role in this child's NTM infection. Person-to-person transmission is rare (5). Predisposing factors for NTM infection in healthy children have not been identified, but it is possible that they have a subtle defect in the interferon-gamma/interleukin-12 pathway (6). Control of NTM infections requires a strong T_H1 response, and it is possible that the predominance of the T_H2 response in our patient (as indicated by her severe eczema) interfered with her response to NTM (7). Conversely, it has been suggested that mycobacterial infections may protect children against symptoms of allergy (8).

Adenitis due to NTM is usually unilateral and most commonly involves the submandibular or anterior superior cervical nodes, suggesting an oral route of entry (9), although any lymph nodes can be involved. The axillary and inguinal involvement in the present case can be explained if MAC entered through eczematous skin lesions on the limbs. Involvement of inguinal lymph nodes usually occurs with involvement of cervical nodes rather than axillary nodes, as in our patient, and for an unknown reason, bilateral inguinal disease is unusual (9). Involvement of multiple lymph nodes is thought to occur with more pathogenic species such as M *haemophilum*, which has been associated with skin lesions, septic arthritis, sinusitis and pneumonia (3).

Patients with NTM lymphadenitis are generally well, without constitutional signs or symptoms (9), and have minimal tenderness in the involved area (10). Over a variable length of time (weeks to months), the overlying skin becomes violaceous as the nodes soften, rupture and drain through a sinus tract to the skin. Sometimes, infection resolves spontaneously or nodes heal without rupturing, but the cosmetic outcome can be poor, with residual fibrosis and calcification. Skin testing is usually positive if NTM antigens are used (9), but there is no commercially available product for such testing. A tuberculin skin test is positive in over 50% of cases (9).

Complete surgical excision of the affected lymph nodes is the traditional treatment for NTM lymphadenitis (10). However, this can require extensive dissection of the neck, can leave scarring and can damage the peripheral facial nerve (2). Incisional drainage is not recommended, because it commonly produces a draining sinus tract or recurrent disease (2). Antimicrobial therapy with a macrolide with or without ethambutol or rifampin is commonly used to obviate or simplify surgical excision. Avoidance of surgery in 30 of 45 cases (67%) with treatment with antimicrobials has been described (4), with the success rate of antimicrobials presumably being increased by early therapy. In another study, clinical response was evident by one month in only approximately 40% of cases successfully treated with antimicrobials alone and by two months in approximately 85% (10), suggesting that it might have been beneficial to give our patient more than a one-month trial of azithromycin. The cosmetic result of medical versus surgical therapy needs to be compared because significant scarring can remain after antimicrobial therapy, especially with spontaneous drainage of the node(s). A randomized trial of antimicrobial versus surgical therapy is ongoing (9).

CONCLUSION

NTM lymphadenitis should be considered in all patients with violaceous, nontender nodes in which exposure to tuberculosis is unlikely.

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