Primary Cervicofacial Nocardiosis due to *Nocardia asteroides* in an Adult Immunocompetent Patient

Monica Corazza[1]*, Luigi Ligrone[1], Marco Libanore[2] and Annarosa Virgili[1]
[1]*Department of Dermatology, University of Ferrara, Via Savonarola 9, 44100 Ferrara, and* [2]*Division of Infectious Disease, S. Anna's Hospital, Ferrara, Italy. *E-mail: czm@unife.it*
Accepted May 8, 2002.

Sir,

The *Nocardia* species are aerobic, filamentous gram-positive bacteria, irregularly acid-fast staining that belong to the order Actinomycetales. Normally, *Nocardia* spp. are soil saprophytes, but *N. asteroides* may be found in the normal flora of the oral cavity and upper respiratory tract. Four species of *Nocardia* are pathogenic in man: *N. asteroides*, *N. brasiliensis*, *N. caviae* and *N. madurae* (1).

N. asteroides is usually the agent of systemic pulmonary infections in immunocompromised hosts, while *N. brasiliensis* is the responsible agent in 74% of all cutaneous manifestations (1, 2).

The skin is generally secondarily involved in disseminated systemic pulmonary diseases, due to haematogenous spread of *N. asteroides*, but it can also be primarily affected.

Primary cutaneous nocardiosis (PCN) accounts for only 5% of all nocardial infections and is caused mainly by *N. brasiliensis* (3, 4). PCN is characterized by numerous clinical manifestations: chronic mycetoma, superficial abscesses and cellulitis and lymphocutaneous variants. The last of these manifestations includes the more common sporotrichoid form (nodules along the lymphatic drainage) and the rarer cervicofacial variant.

We report an unusual case of primary cervicofacial nocardiosis caused by *N. asteroides* in an adult immunocompetent man.

CASE REPORT

A 79-year-old man was admitted to our department because of fever and a necrotic ulcerative lesion of the dorsum of his nose. The borders were vegetant, infiltrated on palpation and a purulent exudate was easily obtained through compression. Numerous sparse tiny pustules and little pus-draining sinuses were peripherically sparse around the ulcerative lesion (Fig. 1). The inflammatory oedema involved the left cheek and the lower eyelid. A hardened swelling of the left mandibular angle and some hard, enlarged, latero-cervical lymph nodes were present.

History revealed that the patient had had a bicycle accident 15 days earlier, causing wounds on his forehead, nose and left cheek. The wounds had healed rapidly with common antiseptic medications.

Blood sample examination revealed white blood cells 13×10^9/l (60% neutrophils, 25% monocytes) and increased ESR (93 mm/h). Paraneoplastic serological markers, immunological investigations and HIV-1/2 serology were negative. Hemocultures were also negative.

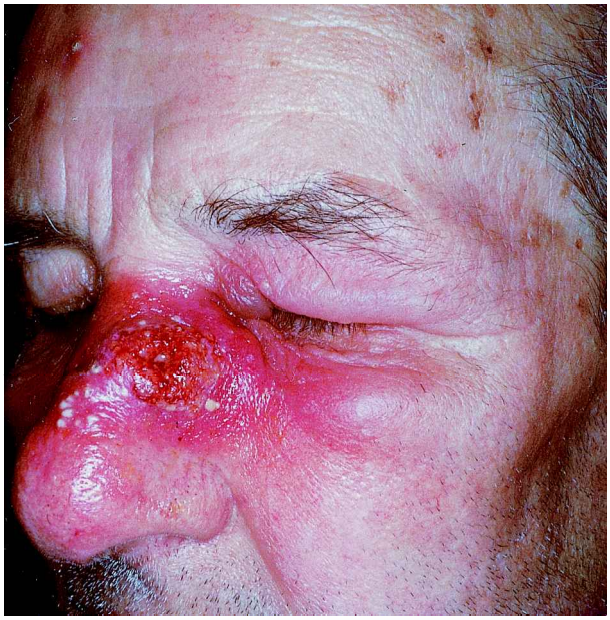

Fig. 1. Inflammatory and ulcerative lesion of the nose.

Skull and chest X-rays were negative while ultrasonography revealed gross hypoecogenic non-homogeneous areas (colliquated lymph nodes) extending to the subfascial area. Sonography also showed numerous colliquated lymph nodes in the right laterocervical region as well as a colliquative involvement of both the parotides and submandibular lymph nodes. Computerized axial tomography of the head confirmed all the data and again showed abscesses of the left masseter muscle and the left submandibular salivary gland.

Histology of a skin biopsy taken from the borders of the ulcer only revealed a dense diffuse inflammatory infiltrate of neutrophils and lymphocytes in the deep dermis, with abscess formation. No fungic elements were observed with PAS staining.

Endovenous therapy with amoxicillin/clavulanic acid 6.6 g/day, amikacin 1 g/day and teicoplanin 800 mg/day was immediately started. A week later a slight improvement of the purulent and inflammatory aspects of the facial lesions was observed but the laterocervical tumefactions had worsened and required surgical drainage. Cultures of purulent exudate from the nose ulcer,

performed on admittance, yielded *N. asteroides* and *Staphylococcus simulans*. Cultures from the exudate from drainage of the lymph nodes and of the biopsy material were negative.

Therapy was immediately switched to endovenous sulphamethoxazole-trimethoprim (400/80 mg), 4 doses twice/day, amikacin 1 g/day and teicoplanin 800 mg/day; this was continued for 2 months, with slow improvement. After this the antibiotics were changed to doxycycline 100 mg b.i.d. and continued for a further 2 months, resulting in complete resolution of the lesions. Follow-up at 1 year was negative.

DISCUSSION

About 70 cases of primary lymphocutaneous nocardiosis have been described in the literature (1, 3–8). Most of the reported cases were sporotrichoid forms on the limbs in otherwise healthy patients caused by *N. brasiliensis*, which is the most virulent species (1, 5–7). *N. asteroids*, a less pathogenic and a true opportunistic agent, is exceptionally the causative agent in immunocompetent patients (4).

Cervicofacial nocardiosis is a distinct subgroup of lymphocutaneous nocardiosis and only sporadic cases have been described in the literature (9–12). It is considered a typical affection in children who develop a pustule in the nasolabial area and successively complain of regional submandibular or cervical lymphadenitis, fever and systemic symptoms. The suppurative evolution of lymphadenopathy may require surgical drainage.

In the paediatric cases reported, cervicofacial nocardiosis was caused by *N. brasiliensis* and only in two cases by, respectively, *N. asteroides* or *N. caviae* (9–12). Cat scratches on the face and the children's playing habits were reported to have caused minor traumas and accidental contamination by soil. The children, aged from 22 months to 7 years, were all cured by antibiotic therapy.

Facial lymphocutaneous diseases have also been reported in 4 healthy adult patients (6, 8, 13, 14) as in our patient.

The prolonged and high dose administration of sulphamethoxazole is the first choice in the treatment of nocardiosis. Other agents that may be of benefit include minocycline, aminoglycosides, third-generation cephalosporins, imipenem and the fluoroquinolones (5, 7, 15). The optimal length of antimicrobial therapy is not known, but long-term therapy is important because of the possibility of recurrence.

REFERENCES

1. Kalb RE, Kaplan MH, Grossman ME. Cutaneous nocardiosis. J Am Acad Dermatol 1985; 13: 125–133.
2. Smego RA, Gallis HA. The clinical spectrum of Nocardia brasiliensis infection in the United States. Rev Infect Dis 1984; 6: 164–180.
3. Shelkovitz-Shilo I, Feinstein A, Trau H, Kaplan B, Sofer E, Schewach-Millet M. Lymphocutaneous nocardiosis due to Nocardia asteroides in a patient with intestinal lymphoma. Int J Dermatol 1992; 31: 178–179.
4. Tsuboi R, Takamori K, Ogawa H, Mikami Y, Arai T. Lymphocutaneous nocardiosis caused by Nocardia asteroides. Arch Dermatol 1986; 122: 1183–1185.
5. Ye Z, Shimomura H, Kudo S, Arao T, Sato Y, Ono T. A case of lymphocutaneous nocardiosis with a review of lymphocutaneous nocardiosis reported in Japan. J Dermatol 1996; 23: 120–124.
6. Naka W, Miyakawa S, Niizeki H, Fukuda T, Mikami Y, Nishikawa T. Unusually located lymphocutaneous nocardiosis caused by Nocardia brasiliensis. Br J Dermatol 1995; 132: 609–613.
7. Paredes BE, Hunger RE, Braathen LR, Brand CU. Cutaneous nocardiosis caused by Nocardia brasiliensis after an insect bite. Dermatology 1999; 198: 159–161.
8. Angelika J, Glander H-J, Haustein UP. Primary cutaneous nocardiosis in a husband and wife. J Am Acad Dermatol 1999; 41: 338–340.
9. Beckmeyer WJ. Nocardiosis. Report of a successfully treated case of cutaneous granuloma. Pediatrics 1959; 23: 33–39.
10. Bates RR, Rifkind D. Nocardia brasiliensis lymphocutaneous syndrome. Am J Dis Child 1971; 121: 246–247.
11. Lampe RM, Baker CJ, Septimus EJ, Wallace RJ. Cervicofacial nocardiosis in children. J Pediatr 1981; 99: 593–595.
12. Law BJ, Marks MI. Pediatric nocardiosis. Pediatrics 1982; 70: 560–565.
13. Sasaky K, Ohara S, Nakakita T, Minamide W. A case of cutaneous nocardiosis of the face. Rinsho Derma (Tokyo) 1992; 34: 821–824.
14. Kimura K, Kuyama M, Mitsugi M. Cutaneous nocardiosis of the face. Rinsho Derma (Tokyo) 1994; 36: 1532–1533.
15. Burgert SJ. Nocardiosis: a clinical review. Infect Dis Clin Practice 1999; 8: 27–32.