Primary Cutaneous Actinomycosis of An Extremity: A Case Report

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Abstract

Primary cutaneous actinomycosis of the extremity is very uncommon and can easily be regarded as ordinary soft tissue infection. Herein, a case of 65-year-old man with primary cutaneous actinomycosis at his lower extremity, probably after direct inoculation of the microorganism from his saliva into a preceding wound in extremity, was reported. He was treated successfully with surgical excision combined with extended period of antimicrobial treatment. (J Intern Med Taiwan 2010; 21: 290-293)

Key Words : Skin; Extremity; Actinomycosis; Saliva

Introduction

Actinomycosis is a chronic and granulomatous suppurative bacterial disease caused by anaerobes Actinomyces, typically involving the areas of cervicofacial, thoracic, abdominal or pelvic areas because of the exclusively endogenous habitat of the bacteria¹. Cutaneous involvement is well documented and it is usually secondary to local extension or probably to hematogenous spreading from various sites. Primary actinomycosis of extremity is very rare with less than 50 case reports in the literature²⁻⁷. Physician should list actinomycosis in the differential diagnoses in patients with chronic non-healing wound in extremities, and particularly give a highly suspicion on the presence of granule-like material from the wound. This reported patient had several well-defined subcutaneous nodules on the right lower extremity and he responded well to surgical excision combined with extended period of antimicrobial treatment. There has been no recurrence during a 1-year period of follow-up. He is probable the first report of primary actinomycosis of extremity in Taiwan. Because of the clinical manifestation of the skin lesion in primary cutaneous actinomycosis is similar to the common skin infection, it makes the diagnostic difficulty and needs a high index of suspicion, especially in patients with a distinct history of wound exposure to saliva.

Case Report

A 65-year-old healthy male was admitted with a 4-year history of recurrent nodular lesions with abscess of his left lower extremity. He recalled that

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the lesion bothered him intermittently and resolved after each treatment with oral antibiotics since the history of traumatic injury 4 years ago when he had applied with his saliva to the lesion for pain relief what he used to do. His past history was not remarkable.

The physical examination revealed several nodular lesions, the biggest one about 2×3 cm in size, with central fluctuation and local heat surrounding by erythematous and swelling changes over his lower third of right lower extremity. Her temperature was 36.4° C, pulse 62 beats/min, respiratory rate 19 breaths/min, and blood pressure 120/78 mmHg. There was no palpable lymphadenopathy.

The laboratory data included a white blood cell count of 6000/ μ L, with neutrophils 55.1%, and lymphocytes 29.2%; hemoglobin of 14.6 g/dL; platelet count of 220 × 10³/ μ L; blood glucose 121 mg/dL; blood urine nitrogen 22 mg/dL; serum creatinine 0.9 mg/dL; sodium 137.9 mmol/L; potassium 3.42 mmol/L; aspartate aminotransferase 45 U/L; alanine aminotransferase 32 U/L; serum albumin 4.2 g/dL; and C-reactive protein 0.25 mg/ dL. The X-ray of lower extremity revealed no significant abnormality. Fine needle aspiration revealed nonspecific inflammation.

Ordinary subcutaneous abscess was impressed initially; hence, antimicrobial therapy with amoxicillin/clavalanate intravenously (1.2 gm per 8 hours) was commenced. On the 2nd hospital day, he underwent surgery of incision and debridement. Under subcutaneous dissection, the pus with many granule-like materials and grey-brown in color was drained out. Tuberculosis or malignancy was impressed by the surgeon. The Gram stain of the pus showed a few neutrophils and gram-positive bacteria. However, the attempt to cultivate the organisms was unsuccessful. The pathological report showed many neutrophils with some colonies consisting of intertwined radiating filaments, capped by eosinophilic hyaline material, creating a sunburst pattern those features are consistent with the diagnosis of actinomycosis (Fig.1). Acid-fast stain showed negative and there was no evidence of malignancy. Hence the diagnosis of actinomycosis was made and amoxicillin/clavalanate was kept. He was discharged on the 6th hospital day to continue the maintenance therapy with doxycycline for 8 weeks. The lesion became cleared and there was no recurrence during the one-year period of follow-up.



Fig. 1.Microscopically, there are many neutrophils with some colonies consisting of intertwined radiating filaments, capped by eosinophilic hyaline material, creating a sunburst pattern.

Discussion

Actinomycosis is a rare infection primarily caused by the Gram-positive, nonspore-forming, anaerobic bacillus Actinomyces spp. It is primarily a commensal microbe found in normal oral cavities, including tonsillar crypts, dental plaques, and carious teeth and female genital tract¹. In addition, animals may also carry this microorganism in different species but cause similar disease^{1.8}. It has never been found in soil, in plants, or in any other object outside the body¹. Most of the infection occurred after traumatic injury that creates an anaerobic conditions predisposing to this microorganism¹, and most commonly associated with other bacterial infections. It is characterized by producing chronic granulomatous, suppurative lesions with abscesses, and drainage sinuses and it easily spreads to adjacent structures and organs. Because of the exclusively endogenous habitat of the etiologic agent, head and neck, thorax, and abdomen are the sites that commonly involved. This microorganism is less virulent than ordinary bacteria; it usually requires a non-intact skin to invade and an anaerobic environment to cause illness. Sulfur granules (grains) containing filamentous or club-shaped structures that are Gram-positive but negative with acid-fast staining are usually found in pus or tissue specimens. However, the identification of organisms alone, in the absence of sulfur granules or an appropriate clinical syndrome, from sputum, bronchial washings, and cervicovaginal secretions is of little significance¹.

Primary actinomycosis of the extremity is very uncommon with less than 50 case reports in the literature²⁻⁷. It has never been described in Taiwan previously; those of actinomycosis reported in Taiwan were usually confined to the common locations, including cervicofacial, thoracic, abdominal, and pelvic regions^{8,9}. This infection usually occurs after direct inoculation after traumatic injury or bites, or less commonly disseminated from primary focus^{1,10,11}. Because of the clinical manifestation of the skin lesion in primary cutaneous actinomycosis is similar to the common skin infection that it makes the diagnostic difficulty and needs a high index of suspicion. Its clinical presentation is usually indolent and has various manifestations, including nodular lesions, subcutaneous abscess, or even mass lesion mimicking tumor¹². Appropriate sampling and histopathological examination are necessary to confirm actinomycosis. Preceding trauma has occurred in most of the patients according to the reports on literature². Lesion may begin in the skin and then spread to contiguous structures, including subcutaneous tissue, muscle, and bone^{1,2,13}.

In this reported case, the diagnosis of actinomycosis is based on the histopathological finding with the typical feature of sulfur granules in pus. Hematogenous seeding is the most likely pathogenesis in patients with primary cutaneous actinomycosis according to the literature review². However, in this case, direct inoculation of Actinomyces spp. from his saliva to his traumatic wound is the presumed pathogenesis. Although culture is definitive, however, it is difficult to cultivate Actinomyces spp. from clinical specimens. In one study conducted by Reiner et al in 1987, the positive culture rate was less than $50\% (17/35)^2$. The reasons for the lower culture rate for Actinomyces spp. could be explained as follows: (1) Actinomyces spp. is very sensitive to a wide variety of antimicrobials, and even a single dose can interfere with their isolation. (2) Strict anaerobic processing and anaerobic growth should be utilized for sampling. (3) Swabs from the lesion that used to do for culture in clinic are usually ineffective for cultivating Actinomyces spp. Therefore, the single most helpful diagnostic maneuver for actinomycosis is to demonstrate sulfur granules in pus or tissue specimens. Treatment of actinomycosis consists of surgical intervention and appropriate antimicrobial therapy. Prognosis is generally excellent. Penicillin is the drug of choice, and tetracycline is the alternative for penicillin-allergic patients^{1,14}. To give initial high dose penicillin and to maintain with oral antimicrobiotics for an extended period for actinomycosis are generally recommended, however, it may depend on the disease severity². This reported patient had focal cutaneous actinomycosis, and he responded well to surgical excision combined with a 9-week course of antimicrobial treatment.

In summary, this case report emphasizes the importance that Actinomyces spp. is a normal

inhabitant of the oral cavity, clinical illness may develop after direct inoculation of this microorganism from saliva into a preceding wound in extremities.

References

- Russo TA. Agents of actinomycosis. In: Mandell GL, Bennett JE, Dolin R, eds. Principles and Practice of Infectious Diseases. 6th ed. Philadelphia: Churchill Livingstone 2005: 2924.
- Reiner SL, Harrelson JM, Miller SE, Hill GB, Gallis HA. Primary actinomycosis of an extremity: a case report and review. Rev Infect Dis 1987; 9: 581-9.
- Sardana K, Mendiratta V, Sharma RC. A suspected case of primary cutaneous actinomycosis on the buttock. J Dermatol 2001; 28: 276-8.
- Wee SH, Chang SN, Shim JY, Chun SL, Park WH. A case of primary cutaneous actinomycosis. J Dermatol 2000; 27: 651-4.
- Okano M. Primary cutaneous actinomycosis of the extremities: a report from Japan. Cutis 1989; 44: 231-3.
- Mah E, Stanley P, McCombe DB. Actinomycosis infection of the finger. Hand Surg 2005; 10: 285-8.
- 7. Fazeli MS, Bateni H. Actinomycosis: A rare soft tissue infection. Dermatol Online J 2003; 11: 18.

- Wang YH, Tsai HC, Lee SJ, et al. Clinical manifestations of actinomycosis in southern Taiwan. J Microbiol Immunol Infect 2007; 40: 487-92.
- Hsu HT, Huang CH, Huang KM, Jang TN. Thoracic actinomycosis mimicking pulmonary tuberculosis: a case report. J Taiwan Emerg Med 2005; 7: 138-45.
- 10.Robinson RA. Actinomycosis of the subcutaneous tissue of the forearm secondary to a human bite. JAMA 1944; 125: 1049.
- 11.Buttas CA, Read SE, Coleman RE, Abravoltch M. Disseminated actinomycosis. Can Med Assoc J 1970; 103: 1069-71.
- Kumar A, Detrisac DA, Krecke CF, Jimenez MC. Actinomycosis of the thigh presenting as a soft-tissue neoplasm. J Infect 1991; 23: 187-90.
- 13.Kargi E, Akduman D, Gungor E, Deren O, Albayrak L, Erdogan B. Primay extremity actinomycosis causing osteomyelitis of the hand. Plast Reconstr Surg 2003: 112: 1495-7.
- Peabody J, Seabury J. Actinomycosis and nocardiosis. A review of basic difference in therapy. Am J Med 1960; 60: 99-115.

原發性下肢放線菌症:一病例報告

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摘要

放線菌症 (actinomycosis) 是一種慢性的化膿性和肉芽腫性傳染病,可能爲局部或全身性。其致病菌是屬於革蘭氏陽性、非抗酸性、厭氣性兼微好氣菌,是口腔中的正常菌叢。臨床上最常感染之部位爲頸面部、胸部,腹部和骨盆腔。原發性肢體放線菌症在臨床上並不多見,其臨床表現與一般細菌感染雷同因而容易被忽略。此報告病例爲一位65歲男性,爲右下肢反覆性化膿性傷口所困擾。經外科手術切除,病理報告證實爲放線菌病。可能的感染來源爲病患習慣性的使用口水塗抹傷口,將口腔內放線菌帶至傷口所致。