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□ Case Report □

Chronic Actinomycosis of the Cervical Lymph Node Simulating a Thyroid Neoplasm

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갑상선종으로 오인된 경부 임파선 만성 방선균증

서영진 · 정 헌 · 진형민 · 조현민 · 원용성 · 김준기
박우배 · 전정수

Actinomycosis in humans is currently a rare disease. Here we report a case of cervicofacial actinomycosis in a 24-year-old man. The patient presented with a painful cervical mass, without symptoms of infection. Clinical features and results of laboratory and imaging studies of the patient suggested a thyroid neoplasm or subacute thyroiditis. Fine needle aspiration cytology failed to yield a definite diagnosis. The pathologic report after a curative operation confirmed the presence of the characteristic sulfur granules in the lymph node just above the left lobe of the thyroid gland. Here we describe this rare case with a review of the typical clinical presentations of actinomycosis on the head and neck, its pathogenesis on the difficulties encountered in the diagnosis and treatment of the disease. **(J Korean Surg Soc 2002;62: 442-445)**

Key Words: Actinomycosis, Cervicofacial, Lymph node
중심 단어: 방선균증, 경안면부, 림프절

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INTRODUCTION

Cervical lymphadenitis is caused by various causes. The causative agents include bacteria, virus, fungus and trauma. And so many disease entities may involve cervical lymph node clinically. Among numerous pathogens, Actinomyces may penetrate directly into the cervical lymph node via minor dental trauma, or diffusely penetrate to the surrounding organs under many conditions. (*1*) Actually cervicofacial Actinomyces comprises about 50% cases of total actinomycotic infections. The incidence of cervicofacial actinomycosis is not high, so it is encountered rarely. The rarity and the absence of characteristic presentations of this infection make the diagnosis extremely perplexing. (*2*) The correct diagnosis can be made after the curative operation, followed by histological examination. Characteristic sulfur granules can help clinicians to confirm the diagnosis. Long-term supplemental antibiotics are mandatory to eradicate the organisms and cure the rare disease.

Here we describe a chronic case of cervicofacial actinomycosis in a man having a left-sided neck mass simulating a thyroid neoplasm. The diagnosis could be confirmed only after surgical removal of the lesion followed by histological confirmation. A long-term supplemental antibiotic treatment after curative resection is mandatory for the eradication of the infection.

CASE REPORT

A 24-year-old man presented with a cervical mass and pain. He had a 1-year history of a left-sided cervical mass, which was not resolved after oral administration of antibiotics, and even enlarging.

On physical examination, a mass measuring 3×3 cm in size was palpated in the left anterior cervical triangle and was thought to originate from the thyroid gland; it was moderately hard, rubbery in consistency, smooth in outline, nontender, and firm. There was no palpable cervical lymphadenopathy.

The blood chemistry and thyroid function tests were normal. Thyroid microsomal antibodies were undetectable. A thyroid scan showed slightly inhomogeneous uptake in the left lobe and

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a suspicious cold nodule in the left upper pole. Ultrasound of the neck confirmed that the palpable mass was an enlarging heterogeneous left thyroid lobe with echo-genic and echo-lucent areas just above the left thyroid lobe (Fig. 1). Fine needle aspiration cytology reports obtained on two different occasions were contradictory. The one suggested a non-functioning thyroid nodule, whereas the other was consistent with a non-specific infection. Computed tomography (CT) imaging of the neck with contrast was consistent with a space-occupying mass arising from the left thyroid lobe with some suspicious abscess formation (Fig. 2). However, radiological and cytologic studies could not give any reliable evidence to lead to a correct diagnosis before operation.

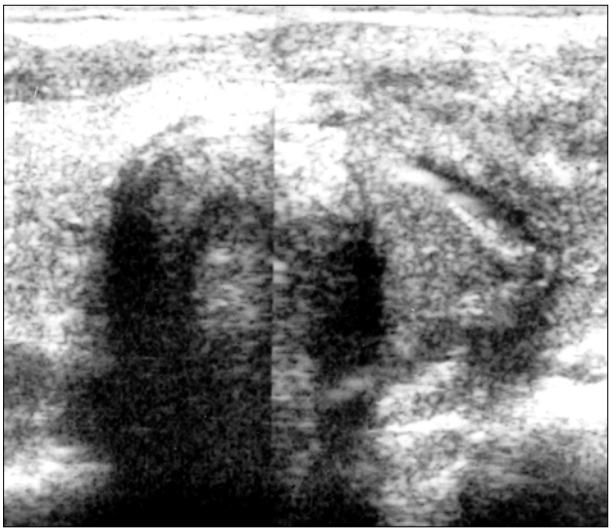

Fig. 1. On ultrasonogram, an ill-defined irregular and heterogeneous area with decreased echogenecity is noted in the left side of the neck, just above the left lobe of the thyroid, with suspicious abscess formation with fistulous tracts.

The patient underwent surgical exploration of the neck, which revealed that the mass consisted of an encapsulated abscess just above the upper pole of the left lobe with an indistinct margin, and was firmly attached posteriorly to the trachea. The platysma and the strap muscles were adherent to the thyroid gland and showed extensive inflammatory infiltration as well as areas of necrosis. The lesion was excised by performing left thyroid lobectomy with isthmectomy. All necrotic tissues were also removed. Grossly thyroid gland looked normal. Microscopic examination revealed the characteristic sulfur gra-

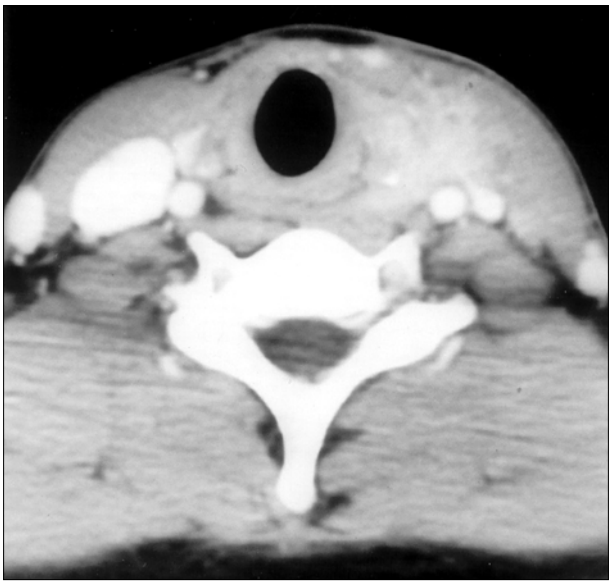

Fig. 2. On computed tomogram, an irregularly shaped enhancing lesion is noted at the left suprathyroidal region, lateral to the left thyroid cartilage. The enhanced lesion also involves the left sternocleidomastoid muscle and upper pole of the left thyroid gland. Multiple small rim-like enhancing portions are noted within the lesion.

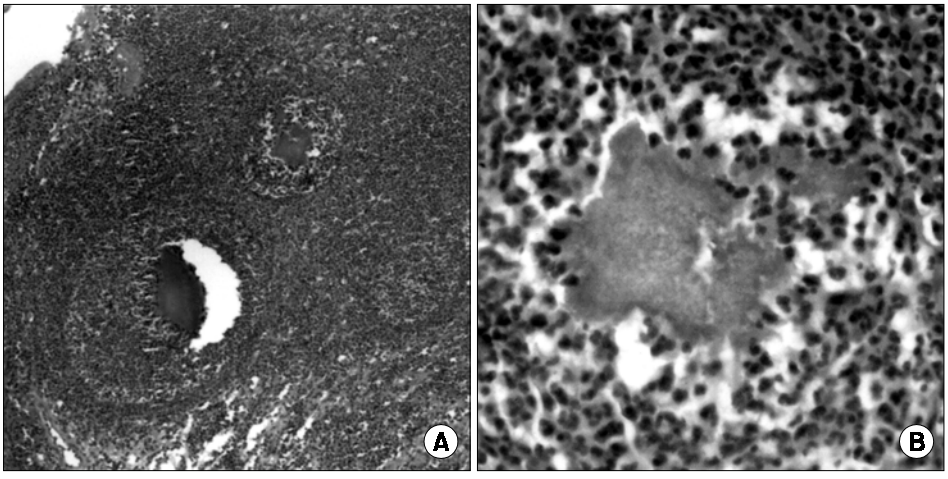

Fig. 3. Histological section showing the characteristic sulfur granule of *Actinomyces* surrounded by acute inflammatory reaction (A. H&E, × 100, B. H&E, ×200).

nules of *Actinomyces* surrounded by acute and chronic inflammatory reaction in the lymph node (Fig. 3). After the diagnosis was made, he received intravenous clindamycin (300 mg every 6 hours for two weeks), which was followed by oral doxycycline (100 mg twice daily for six months).

The patient is now asymptomatic in a complete clinical remission status.

DISCUSSION

Thyroiditis is caused by a variety of pathological processes including autoimmune disease, trauma, radiation, and infection. However, thyroiditis caused by bacterial or fungal infection is rare. Bacterial thyroiditis is a very uncommon disease caused most often by gram-positive cocci such as *Staphylococcus aureus*, *Streptococcus pyogenes*, and *Streptococcus pneumoniae*. *Actinomyces* species are one of the causative agents for infective thyroiditis, but are encountered extremely rarely.

The first case of thyroid actinomycosis was reported in 1894. (*1*) The rarity and the absence of characteristic presentations of this infection make the diagnosis extremely perplexing. (*2*)

Actinomycosis is an indolent, slowly progressive infection caused by anaerobic or microaerophilic bacteria, primarily of the genus *Actinomyces*. *Actinomyces* are closely related to mycobacteria and taxonomically fall between true bacteria and fungi. They were a common source of human morbidity before the modern antibiotic era. *Actinomyces* are gram-positive, branching, filamentous, anaerobic-to-microaerophilic microorganism. In humans, they are normal symbiotic inhabitants of the oral cavity of healthy hosts. Thus, actinomycetal infections can be caused by the spread of the organism from the mouth to the surroundings under various conditions. Actinomycosis usually involves the cervicofacial area (50% of total cases), the thorax and the abdomen is less common than cervicofacial area. (*3-6*) Thyroid involvement is rare in cervicofacial actinomycosis.

The pathogenesis of cervicofacial actinomycosis is poorly understood. Because the organism cannot penetrate the mucosal barrier on its own, an interruption of oral mucosal integrity is perhaps the most important permissive factor for actinomyces invasion. Poor oral hygiene, periodontal disease or gingivitis, as well as mucosal trauma or manipulations during minor dental surgery are the usual predisposing factors. Because actinomycosis spreads by direct extension rather than by lymphatics or hematogenously, involvement of the thyroid gland is not easily explained. The rarity of involvement of the thyroid gland by *Actinomyces* led some authors to suggest that this gland might be unusually resistant to bacterial infections, especially actinomycosis. Although there is a general agreement about the usual portal of entry in actinomycosis, it is less clear by which specific routes the organisms reach the thyroid gland. In primary thyroid infections, hematogenous dissemination seems most probable. Presence of the thyroglossal duct or a piriform sinus fistula potentially provides communication between the oral cavity and the thyroid parenchyma. Persistence of a patent thyroglossal duct has been previously implicated in the pathogenesis of thyroid actinomycosis. (*7*) In some reports, the involvement of the thyroid was a part of generalized infection (actinomycotic pyaemia) or it was due to contiguous growth following an actinomycosis of the soft tissues of the head and neck.

Clinical manifestations of the actinomycosis of the head and neck are presented in two ways. A firm, hard, slightly painful and slowly increasing swollen mass may be found at the point where the lower limit of the mandible meets the facial blood vessels and may be accompanied with development of draining fistulous tracts penetrating up to the skin in a severe form. Otherwise, it may simulate an acute infection in the submandibular area, at the angle of the mandible, in the parotid region, or in the neck. In the latter acute form, there is usually suppuration and abscess formation beneath the skin with subcutaneous edema and inflammatory changes, such as wheal or redness with local heat, in the surrounding tissues. In this case, however, the patient lacked any of these signs. Lymphadenopathy is an uncommon manifestation unless contiguous nodes are involved directly as shown in this case. (*4*)

The clinical diagnosis of actinomycosis is frequently overlooked and is delayed for a long time, due to its rarity and the absence of common clinical presentations of infection. The diagnostic clue is the characteristic sulfur granules, which is the dense knotted colonies of coalescing organisms and this gross morphology is due to an acute and chronic inflammatory response with extensive fibrosis, with typical branching pleomorphic rods in tissue material. However, sulfur granules are not pathognomonic of actinomycosis because *Nocardia* species can also form sulfur granules, although it is easily distinguished by positive acid-fast staining and aerobic growth. Thus, a positive culture is required to confirm the histological diagnosis. Macroscopically, actinomycosis is characterized by a typical single abscess or multiple abscesses with a hard fibrous wall and by the formation of fistulous tracts that extend even to the skin or the surrounding tissues, especially in the cases with thoracic or abdominal involvement. (*3,4,6,8,9*)

After the first report in 1894, about 15 additional cases of

the thyroid actinomycosis have been described. (2) A postoperative infection after a thyroidectomy incision has also been reported, while passage of the *Actinomyces* through a persistent thyroglossal duct has also been suggested. (*3,7,8*)

The disease is usually suspected at surgery. In an attempt to diagnose cervicofacial actinomycosis preoperatively, Pollock et al. (*10*) used a fine needle aspiration technique. Arfeen et al. (*11*) also succeeded in diagnosing a thyroidal actinomycosis with a fine needle biopsy performed under ultrasound guidance. However, we failed to locate the sulfur granules in the aspiration specimens, indicating that the fine needle aspiration may not always be helpful.

In the present case, none of clinical findings suggested the diagnosis of thyroid actinomycosis. The disease appeared as a firm, not indurate minimally painful swelling mass in the absence of enlarged regional lymph nodes or elevated body temperature indicative of an apparent clinical infection. These findings, together with the normal leukocyte count and erythrocyte sedimentation rate, were not consistent with an infectious process. Furthermore, the fine needle aspiration cytology results were inconclusive, while the CT scan images were suggestive much of a neoplasm. The finding, however, of an abscess formation at surgery and the identification of *Actinomyces* by histological examination confirmed the diagnosis of actinomycosis. However, the source of infection in this patient remains unknown.

Recurrence is the rule in actinomycosis following surgery alone, and therefore, postoperative antibiotic supplementation is mandatory for successful treatment. The choice of antibiotic for actinomycetal infections has been penicillin. In case of penicillin allergy, tetracyclines, erythromycin, or clindamycin can be chosen as an alternative. The treatment may fail either because of a truncated antibiotic prescription or because of patient's poor compliance. It is crucial that the antibiotics should be maintained for six to twelve months, whatever the agents should be. Subtotal thyroidectomy should be considered, if a recurrence occurs in spite of the thorough medical treatment, to eradicate the infection. (3,5,8)

We suggest that the actinomycosis should be suspected during the diagnostic work-up of a thyroid mass or a soft tissue mass around the thyroid glands, especially there is evidence of inflammation around the mass, and in the cases with enlarged lymph nodes, even though there are no local signs of an infectious process. The definitive diagnosis of actinomycosis usually requires operation with pathologic confirmation. (*12*)

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