대한외과학회지 : 제 63 권 제 3 호
Vol. 63, No. 3, September, 2002

□ Case Report □

Actionmycotic Thyroiditis in a Child

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소아에서의 원발성 갑상선 방선균증 1예

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The authors report a rare case of actinomycosis, involving the thyroid gland in a 9-year-old female, presented with a hard, non-tender, non-inflammatory left neck mass of 4 weeks duration. On physical examination a firm, non-tender mass within the left thyroid gland was found; and the laboratory and imaging studies strongly suggested a thyroid neoplasm. After a left subtotal thyroidectomy, a gross sulfur granule was founded in the excised lesion. Histological examination confirmed actinomycotic thyroiditis. Actinomyces species are a very rare cause of infective thyroiditis, with difficulties in their diagnosis and treatment. The authors reviewed its pathogenesis and treatment. **(J Korean Surg Soc 2002;63:262-266)**

Key Words: Actinomycosis, Thyroiditis
중심 단어: 방선균증, 갑상선염

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INTRODUCTION

Thyroiditis is caused by a variety of pathological processes including autoimmune disease, trauma, radiation and infection. However, thyroiditis caused by bacterial or fungal infection of the thyroid gland is rare. Bacterial thyroiditis is a very uncommon disease caused most often by gram-positive cocci such as Staphylococcus aureus, Streptococcus pyogens, and Streptococcus pneumoniae. Actinomyces species are extremely rare causes of infective thyroiditis. The first well-documented case of thyroid actinomycosis was described as early as 1894, with only about 15 cases having been reported since then. (*1-3*) The authors report a rare case of actinomycotic thyroiditis occurring in the pediatric age group, the correct diagnosis of which only became apparent after the surgical excision of the lesion and its histological examination.

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Received June 26, 2002, Accepted July 16, 2002

CASE REPORT

A 9-year-old-female was referred for assessment to our Department of Surgery for an asymptomatic neck mass of 4 weeks duration that did not appear as a inflammatory lesion at any time. She denied any symptoms such as fever, malaise, easy fatigue, sore throat or local tenderness. She gave a history of viral meningitis treated three years ago. Physical examination revealed an afebrile child in a good general condition. A mass measuring 2×2 cm in size was palpated in the mid-portion of left lobe of thyroid gland; it was moderately hard in consistency, smooth in outline, non-tender, and moved with swallowing. There was no palpable cervical lymphadenopathy. The intraoral examination revealed good dentition without canine teeth and periodontitis. The remainder of the examination showed normal results with no signs suggestive of either hyperthyroidism or hypothyroidism. The white blood cell (WBC) count was 7,300/ mm^3, with a normal differential count. Hemoglobin was measured at 12.7 gm/ dl. The blood chemistry and urinalysis were within normal range.

The thyroid function tests revealed a normal thyroid level with a T4 of 10.80 nCug/ dl (normal range 4.700 12.500), a T3 of 177.00 Cug/ dl (normal range 78.00 0 182.0), and a TSH of 3.00 uIU/ ml (normal range 0.170 4.050 uIU/ ml). Thyroid microsomal and thyroglobulin antibodies were not detected. Ultrasound of the thyroid confirmed that the palpable mass was an about 2 cm sized well-defined mass involving the mid portion of the left thyroid gland (Fig. 1). The mass presented mottled internal calcifications and peripheral layered solid portions. A 99metastable-technetium thyroid scan showed that a photon defect was noted in the otherwise normal looking left lobe of thyroid (Fig. 2). Fine needle aspiration cytology biopsies obtained on two different occasions were insufficient for a proper diagnosis. A presumptive diagnosis of a thyroid neoplasm was made. The patient underwent surgical exploration of the thyroid, which revealed that the mass was well-capsulated within the left thyroid lobe, not adherent to adjacent strap muscles and trachea. The lesion was excised by performing left subtotal thyroidectomy. The cut section findings of the mass showed a well-capsulated cavity without necrotic and purulent material (Fig. 3). The most

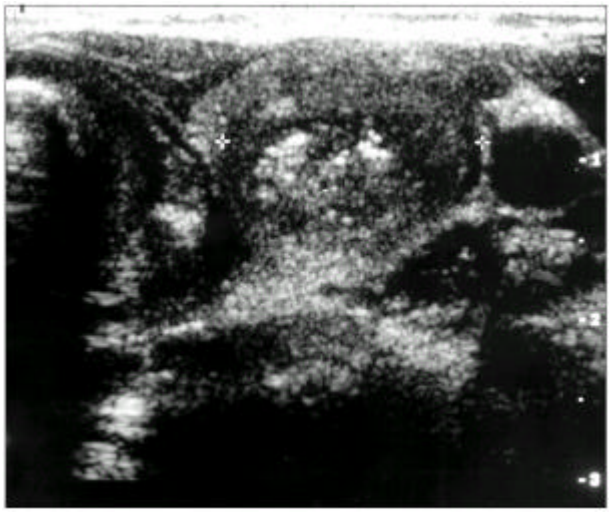

Fig. 1. Ultrasound of the neck demonstrates a solid mass in the mid-portion of the left lobe of the thyroid gland.

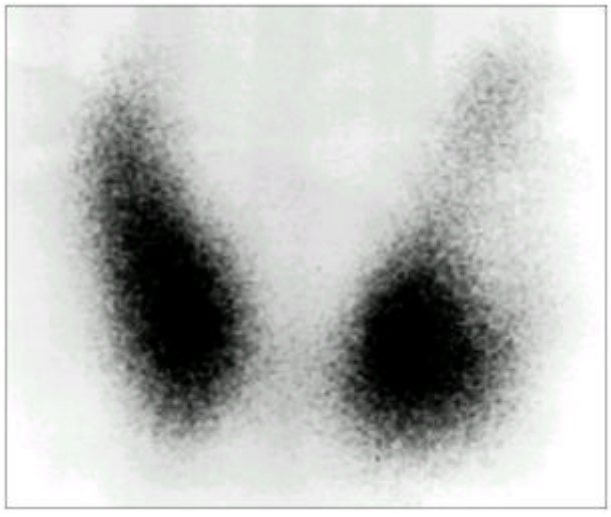

Fig. 2. A 99metastable-technetium thyroid scan showed that a photon defect was noted in the otherwise normal looking left lobe of thyroid.

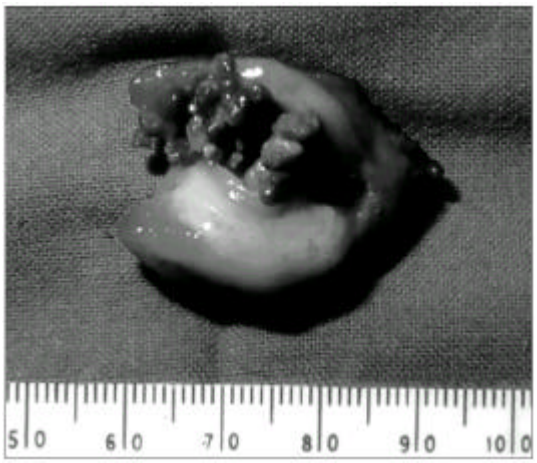

Fig. 3. The cut surface of specimen shows dark brownish sulfur granule in it.

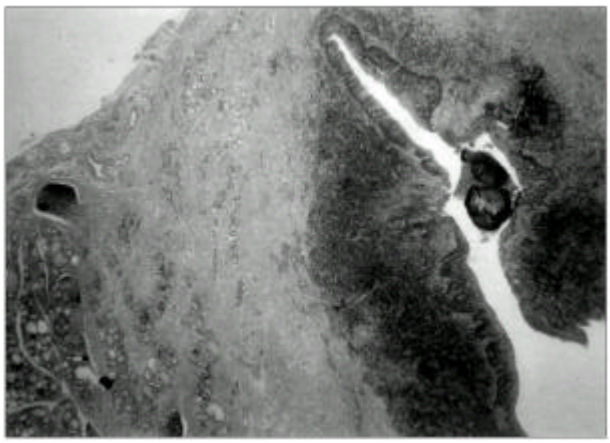

Fig. 4. There is an irregular sinus tract in the thyroid, with some colonies of microorganism (hematoxylin-eosin, ×100).

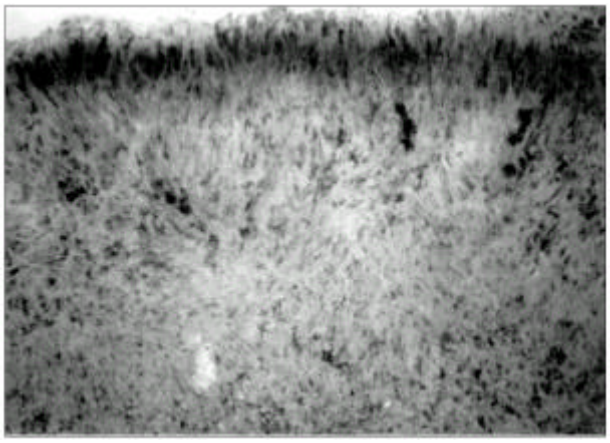

Fig. 5. The edge of the organism showing 1 μm diameter filaments. The peripheral filaments often terminate in a club (hematoxylin-eosin, ×400).

common components were collections of bright green particles measuring about 0.2 cm in diameter each. Culture of the content was not done. Histological examination of these particles showed characteristic sulfur granules of Actinomyces composed of filamentous microorganisms surrounded by an acute inflammatory reaction (Fig. 4, 5). She was discharged at third hospital day postoperatively without any problems. The patient has been received oral penicillin preparations. The patient has remained asymptomatic through weekly follow ups.

DISCUSSION

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 exist taxonomically between the true bacteria and fungi, and were a common source of human morbidity before the modern antibiotic era. These gram-positive bacteria are filamentous (0.5 to 1.0 mm in diameter) with branching and are non-acid fast, with a tendency to break up into coccobacilli. Actinomyces may colonize in the oral cavity, colon, and vagina. The species most frequently involved in humans are Actinomyces israelii, naeslundii, odontolyticus, and viscosus. Actinomycosis is observed throughout the world, and its prevalence is unrelated to climate, occupation, race or age. Infection occurs throughout life, with a peak incidence in the middle decades. Males have a threefold higher incidence of infection. (*4*) Actinomycosis generally presents as a focal inflammatory process involving soft tissue, characterized by an indolent course, local induration, minimal tenderness and few constitutional symptoms.

Actinomyces may be categorized into the head and neck type, the thoracic type, the abdominal type and the disseminated type, according to the infection origin. Weese et al. (*5*) have reported that amongst 57 reported cases, the head and neck type accounted for more than half with 28 cases, with the abdominal type, the thoracic type and others ranking in order of frequency. Actinomycotic thyroiditis may be classified as the head and neck type of Actinomyces. We diagnosed a mass in the left anterior neck portion of a nine year old female patients a thyroid tumor and conducted a left subtotal lobectomy accordingly. The pathological studies of the incised tissue showed the typical characteristics of Actinomycotic thyroiditis such as chronic inflammation, necrosis and the appearance of sulfur granules. Before surgical resection, the mass presented itself as a woody hard area that was relatively painless with applied pressure, traits that let us to initially believe it to be a thyroid tumor.

Cervicofacial actinomycosis comprises 50 to 60% of reported cases. Cervicofacial actinomycosis frequently presents as a firm, indurated, and minimally painful swelling. These features together with the often absent regional lymphadenopathy, lack of pyrexia, and normal white cell count are suggestive of a proliferative process rather than an infection. So Actinomycosis is often mistaken for a neoplasm. (*2*) With time, spontaneous sinus tract development with drainage of sulfur granules can be seen. (*6*)

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 usual predisposing factors. (*7*)

The clinical diagnosis of actinomycosis is frequently missed with a long diagnostic delay. The dia-

gnostic clue is the finding of sulfur granules with typical branching pleomorphic rods in tissue sections or drainage material. However, sulfur granules are not pathognomic of actinomycois; Nocardia species can also form sulfur granules, although it is easily distinguished by positive acid-fast staining or aerobic culture. It is important to confirm the histologic diagnosis by culture studies. (*7*) All too often, the first mention of actinomycosis is by the pathologist after surgery has been performed. Both fine-needle aspiration and biopsy are being used successfully to obtain clinical material for diagnosis. Immunofluorescence testing for A. israelii, A. naeslundii, and P. propionicum has become a useful diagnositc alternative.

Cervicofacial actinomycosis occurs most frequently at an oral, cervical, or facial site. The submandibular space is the most common site for cervicofacial involvement. (*8*) According to Leers et al. (*1*), the first reported case of thyroidal actinomycosis dates back to 1894. About 15 additional cases have been described since then. (*9*) Because actinomycosis spreads by the direct extension and generally not by lymphatics or vessels, invlovement of the thyroid gland is less easily explained. The rarity with the thyroiditis by Actinomyces led some authors to suggest that this gland might be unusually resistant to bacterial infections, and the actinomycosis in particular. Although there is a general agreement about the usual portal of entry in actimomycosis infections, it is less clear by which specific routes the organisms reach the thyroid. In primary thyroid infections, hematogenous dissemination seems most probable. Presence of the thyroglossal duct or a piriform sinus fistula potentially provides a communication between the oral cavity and the thyroid parenchyma. Persistence of a patent thyroglossal duct previously has been implicated in the pathogenesis of thyroid actinomycosis. (*10*) In some reports, the involvement of the thyroid was a part of a generalized infection (actinomycotic pyaemia) or a consequence of a contiguous growth following an actinomycosis of the soft tissues of the head and neck. A post-operative infection from a thyroidectomy incision has been reported. (*1,2,9*) In an attempt to diagnose cervicofacial or actinofacial actinomycois pre-operatively, Pollock et al. (*11*) used fine needle aspiration techniques. According to these authors, the aspiration specimens were adequate for morphogic studies, with sulfur granules remaining intact and suitable for bacteriologic isolation. Arfeen et al. (*3*) also succeeded in diagnosing a thyroidal actinomycosis with a fine needle biopsy performed under ultrasound guidance. But, in an our case, FNAB revealed insufficient results even in two trials.

As with other forms of infection, thyroid actinomycosis responds best to combined medical-surgical therapy. Penicillin G is the drug of choice for treating an infection caused by the species of Actinomyces. It should be given in high dosage over a prolonged period, because the infection has a tendency to recur, presumably because antibiotic penetration to areas of fibrosis and necrosis into sulfur granules may be poor. Long-term (3 to 12 months) therapy is indicated. Otherwise there are no specific guidelines regarding the treatment of actinomycosis involving the thyroid gland. There is a minor evidence concerning the acquisition of resistance to penicillin G by Actinomyces during prolonged therapy. Alternative first-line antibiotics for treating Actinomyces infections include tetracycline, erythromycin, and clindamycin. First-generation cephalosporins ceftriaxone, and imipenem have also been employed successfully.(*12*)

Neck masses are common in childhood, and they can present a source of alarm among parents and a diagnostic challenge for the clinician. The differential diagnosis includes congenital, inflammatory and neoplastic lesions. Pediatric cervicofacial actinomycosis is an exceedingly uncommon entity and thyroid involvement is even more unusual. We experienced one case of actinomycotic thyroiditis in a 9-year-old child after surgical excision. There was nothing to suggest the diagnosis of thyroid actinomycosis in physical, laboratory, and imaging studies. Fine needle aspiration did not prove to be helpful in this case. Actinomycosis should be considered in the differential diagnosis of a neck mass even in childhood.

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