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CERVICOFACIAL ACTINOMYCOSIS: REPORT OF TWO CASES

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ABSTRACT

This article presents two cases of actinomycosis. Case 1 was a 39-year-old man who was first seen with the chief complaint of swelling around the left submandibular region. Case 2 was a 40-year-old woman who was first seen with the chief complaint of mass formation around the left buccal area. Both cases were initially diagnosed as malignant tumors and were later histopathologically interpreted as actinomycosis because of the presence of sulfur granules.

Key Words: Actinomycosis, Sulfur granules

INTRODUCTION

Recently, the incidence of actinomycosis in humans has greatly decreased. However, we seldom encounter a lesion with typical manifestions of actinomycosis. This bacterial disease presents many clinical findings similar to certain malignant tumors. In the present study, two cases of actinomycosis are discussed that were initially diagnosed as malignant tumors and then histopathologically interpreted as actinomycosis because of the presence of sulfur granules in the tissue sections from biopsy or operation materials.

Case 1

The patient was a 39-year-old man who was first seen on October 5, 1984 with the chief complaint of swelling around the left submandibular region. About two months earlier, he had experienced the extraction of an embedded tooth ($\boxed{8}$) followed by swelling of the left submandibular region (Fig. 1). On examination, the swelling extended from the left and right submandibular regions to below the thyroid cartilage. The lesion showed diffuse swelling accompanying an elastic, hard induration. The extraction wound was healing well. No spontaneous or oppressive pain or trismus could be detected. Although the left submandibular lymph nodes were not palpable due to the swelling, a movable lymph node swelling measuring 1.5×1.0 cm in diameter was observed in the right submandibular region. There was no particular finding in the patient's family histroy. All other laboratory data, excluding CRP value (7.1 mg/dl) were within normal limits. A CT scan showed an isodense mass in an area ranging from the submandibular gland to the hyoid bone (Fig. 2). A gallium scintigram indicated abnormal uptake in the left submandibular area, while no abnormal finding was detected on the sialoscintigram and lymph node scintigram nor in the results obtained by sialography (Fig. 3). The clinical findings suggested a malignant tumor arising in the submandibular region.

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Fig. 1. Clinical photograph taken at pretreatment time. The swelling extended from the left and right submandibular regions to below the thyroid cartilage (arrow).

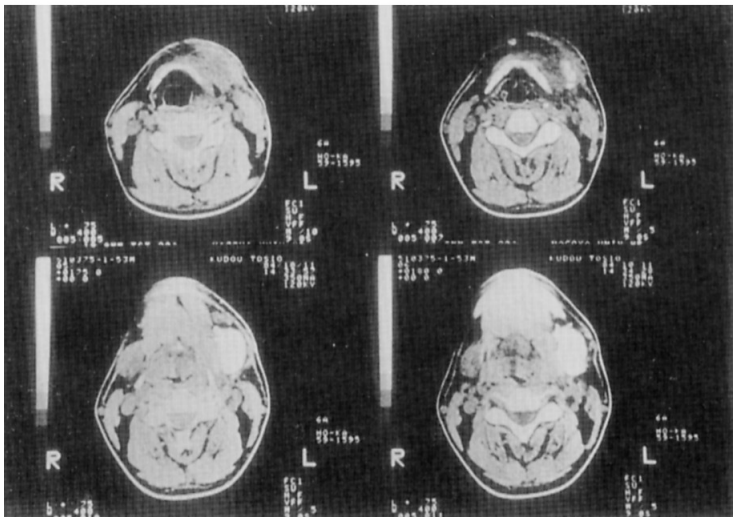

Fig. 2. A CT scan showing the isodense mass present in the submandibular gland and the hyoid bone area.

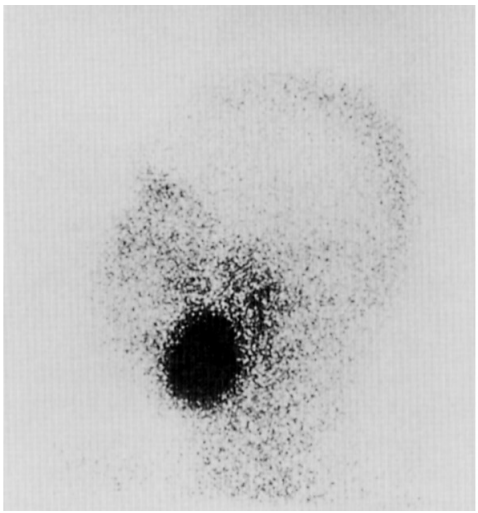

Fig. 3. Gallium scintigram showing abnormal level of Ga uptake in the left submandibular gland.

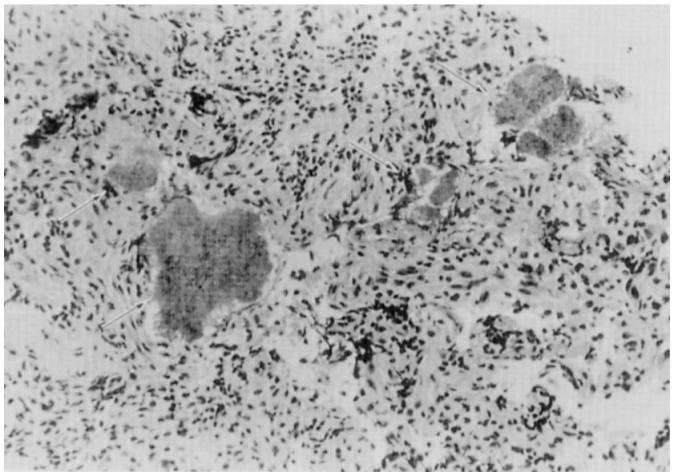

Fig. 4. Photomicrograph showing sulfur granules (arrow). (Hematoxylin and eosin. Original magnification, ×20)

An incisional biopsy was made under local anesthesia. The biopsy material was histopathologically interpreted as an inflammatory lesion with massive inflammatory cell infiltration and sulfur granules (Fig. 4). Thus, a diagnosis of actinomycosis was made on the basis of these findings. Ampicillin was administered (2 g/day) intravenously, in combination with hyperbaric oxygen therapy (OHP). About 10 days after the start of therapy, the induration present in this lesion softened and then disappeared completely. There is no evidence of recurrent lesion.

Case 2

The patient was a 40-year-old woman who was first seen on December 12, 1985 with the chief complaint of mass formation around the left buccal area. The patient noticed a gradually growing, painless mass the size of the tip of the little finger around the left buccal area. On oral examination, a hard, slightly elastic, movable and painless mass the size of the tip of the thumb was observed in the left buccal area. The submandibular lymph nodes on both sides were palpable, painless, and movable. In the mucosa overlying the left buccal area just below the parotid papilla, the mass could be detected by palpation. Discoloration or ulceration of the overlying mucosa was not evident. There was no particular finding in the patient's past and family histories. All other laboratory data, excluding white blood cell count (12800/μl) and total cholesterol value (296 mg/dl), were with in normal limits.

Surgical excision of the mass was conducted under general anesthesia. Because the mass was embedded in the buccal muscles and adipose tissues, an enbloc resection was performed. The resected mass was 20 mm×20 mm×15 mm in size and weighed 1.5 gr. The cross section was gray-black with a reddish tinge. There were no findings that suggested a tumor. Thus nonspecific inflammation was suspected (Fig. 5). The tissue sections revealed inflammatory cell infiltration such as leukocytes or lymphocytes as well as foamy cells and sulfur granules (Fig. 6). Thus, this lesion was interpreted as being actinomycosis. The patient was given 1.2 million units of benzylpenicillin intravenously and was discharged one month after the operation. Chemotherapy was continued on an out-patient basis. As of November 8, 1990, the surgical wound was healing without evidence of recurrent lesion in the area of excision.

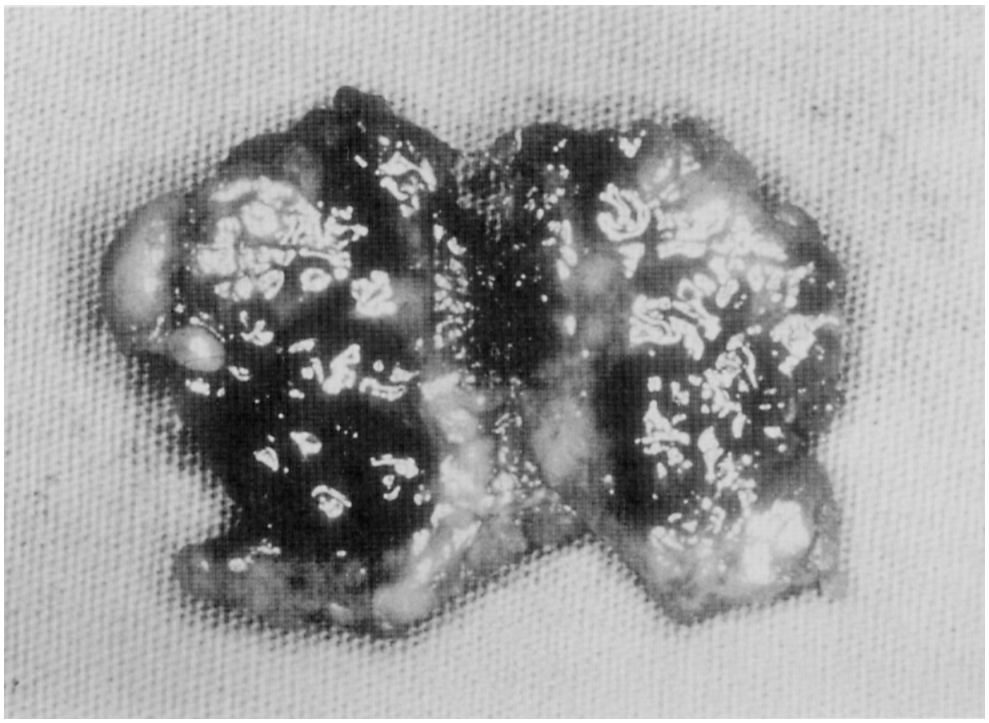

Fig. 5. Photograph showing the cross section of the removed mass.

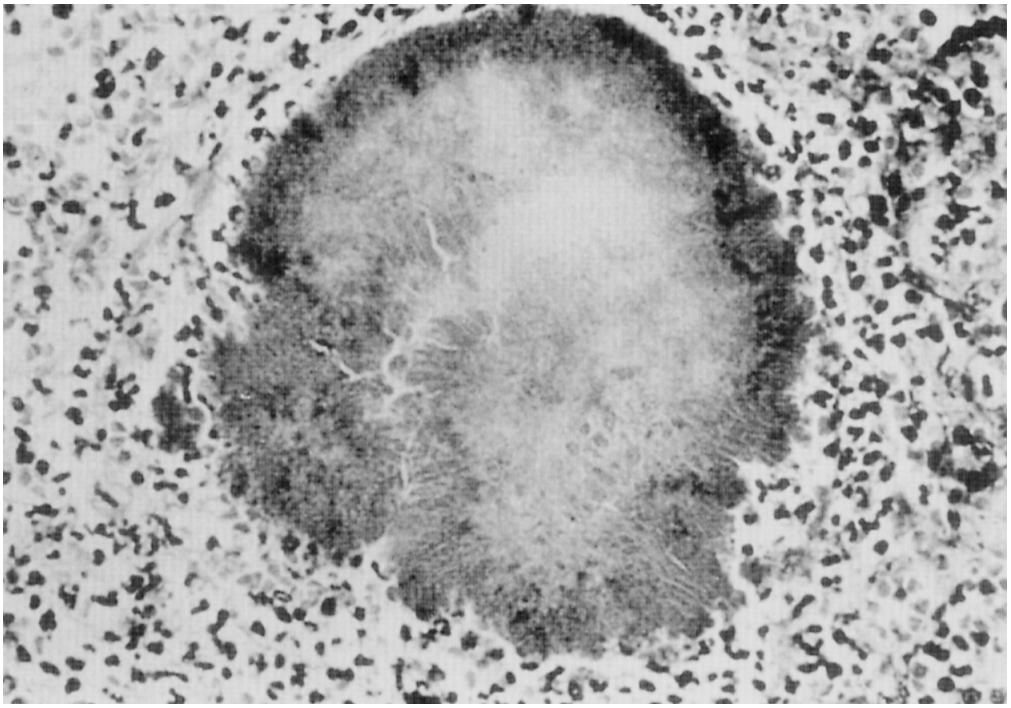

Fig. 6. Photomicrograph showing typical sulfur granules. (Hematoxylin and eosin. Original magnification, ×40)

DISCUSSION AND CONCLUSION

Cervicofacial actinomycosis caused by the infection of Actinomyces israelii and usually occuring in the oral cavity, proceeds to the chronic disease processes and is often insensitive to conventional therapy. The occurrence of actinomycosis in the oral region is usually caused by operation or odontogenic infection. In the two cases reported in the present paper, tooth extraction was followed by the appearance of cervicofacial actinomycosis in the first case, while the cause of disease remains to be proven in the second case. A malignant tumor was initially suspected in both cases because of the absence of evident signs of infection and the presence of a rapidly growing tumor-like mass. However, these cases were finally diagnosed as actinomycosis. Along with developments in antibiotics, there have been considerable modifications in the clinical course of this infection. Namely, the typical clinical signs and symptoms of cervicofacial actinomycosis previously described in the textbooks, such as woody fibrosis (characteristic induration), multiple sinus tracts, trismus and presence of sulfur granules, are often absent. Thus, we feel that it is difficult to diagnose actinomycosis clinically. Brown reported that only 19 (10.5%) of 181 cases of actinomycosis were diagnosed as actinomycosis at the first examination.[1)]

In actinomycosis of the cervicofacial region, although there are acute and chronic stages, the clinical course varies and patients may display many combinations of both acute and chronic features. Asada et al.[2)] have reported that in the acute stage of actinomycosis, Streptococcus and Peptostreptococcus infections are most commonly found; while in the chronic stage, anaerobic organisms are most frequently found. Low virulent anaerobic bacteria such as Actinomyces may

thus function as important pathogenic organisms. In the chronic stage of actinomycosis highly virulent organisms such as Streptococcus destroy tissues, enlarge the area of infection and create conditions that promote the growth of Actinomyces. Thus, the characteristic signs and symptoms of actinomycosis develop in the diseased sites. In the acute stage, chemotherapy prevents development of the conditions; therefore, woody fibrosis, multiple sinus tracts and sulfur granules do not appear at this stage of the disease. Both cases in the present papar were clinically interpreted as being malignant tumors. However, because lymphocytes and plasma cells as well as foamy cells and sulfur granules were found in the tissue sections, but no neoplastic cells, a diagnosis of actinomycosis was made. The clinical findings of this rare bacterial disease are similar to those of certain malignant tumors. Diagnosis was not initially possible due to inadequate examinations using a thin-needle aspiration biopsy, a well-known technique for diagnosing neoplastic disease.[3)]

Surgery and antibiotic therapy (penicillin) were conducted in the second case, and ampicillin in combination with hyperbaric oxygen was administered in the first case. Surgical incision is important to ensure that adequate amounts of antibiotics are actually delivered to the site of infection. Usually high doses of penicillin, administered over a period of weeks to months, are preferred.[4)] Clindamycin should be considered the drug of choice for patients allergic to penicillin.[5)] Erythromycin, tetracycline and lincomycin are other options for the treatment of actinomycosis. Oral cephalexin and semisynthetic penicillins oxacillin and dicloxacillin are less effective in vitro and should not be used, if possible.[6)] Hyperbaric oxygen therapy enhances the oxidation-reduction potential of tissues to levels at which harmful anaerobic organisms are inhibited or killed and also promotes the rapid healing of surgical debridement.[7)]

At the time of diagnosis, portal of entry, patient's present history and length and type of treatment should be carefully considered. A biopsy or identification of pathologic organisms should also be performed. Special care must also be taken as to the choice of antibiotics to be administered.

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