A Case of Actinomycosis of the Minor Salivary Gland in the Buccal Region

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Abstract: We report a case of actinomycosis arising in the minor salivary gland in the buccal region. A 71-year-old male presented with a swelling in the left buccal region. The clinical diagnosis was minor salivary gland tumor in the buccal mucosa. Under local anesthesia, the lesion was excised. Histopathological examination showed basophilic amorphous masses of *Actinomyces* in the dilated excretory duct with squamous metaplasia. A final diagnosis of actinomycosis was made. Its portal of entry was thought to be a disruption of the mucosal barrier after trauma due to maladaptation of dentures. There was no sign of recurrence after the surgery.

Key words: actinomycosis, buccal salivary gland, gram staining, Grocott staining

Introduction

Actinomycosis is a granulomatous inflammatory disease which shows slow progress and often arises in the cervico-maxillofacial region. Multiple abscesses, board-like induration and trismus are typical signs of actinomycosis. But recently, such typical symptoms have decreased in frequency and it has become difficult to diagnose the lesion clinically\(^1\)–\(^3\). The causative organism of actinomycosis is mainly *Actinomyces israelli* which is a gram-positive anaerobic bacterium. In the maxillofacial region, the sites of predilection of actinomycosis are the mandible (80%) and maxilla (10%). *Actinomyces* infection in the minor salivary gland is uncommon (1%)\(^4\). We report a case of actinomycosis of the minor salivary gland in the buccal region.

Case Report

A 71-year-old male was referred to our department with the chief complaint of a mass at the left buccal region in April 2006. The symptoms were first noticed at six months prior to the referral by himself, but he did not seek medical attention because there was no pain and no change in the size of the mass. His medical history included well controlled diabetes. Intraoral examination revealed that a mass, measuring 9 × 8 mm, was located in the left buccal region corresponding to the lower left first premolar area (Fig. 1). The mass was round and movable, elastic and hard, and the surface of the mucosa showed normal color. It was not tender. He was dentulous and wore a partial denture at the left upper jaw. His oral hygiene condition was well controlled. The regional lymph node was not significantly affected. A diagnosis of minor salivary gland tumor or inflammation such as lymphadenitis was suspected clinically. The mass was small. At first a benign minor salivary gland tumor was suspected, so he was observed for one month without carry-
ing out any imaging examination. One month later, the mass had slightly increased in size to 14 × 8 mm. He complained of a yellowish white pus discharge from the mass, so we suspected a secondary infection of the minor salivary gland tumor. Under local anesthesia, we excised the mass surgically. The mass was multilobular and solid. We administered cefem-type antibiotics for three days.

Histopathological examination revealed that there was a dilated excretory duct with squamous metaplasia. Suppurative exudates and basophilic amorphous masses of bacterial colonies could be seen in a duct of the minor salivary gland, but not in the submucosal tissues surrounding the minor salivary gland (Fig. 2A, B). Gram staining showed gram-positive organisms (Fig. 3A). Grocott staining (Gomori’s methenamin-silver staining) showed black-colored filamentous hyphae forming a radially arranged complex network (Fig. 3B). In the periphery of the duct, there was fibrous capsule-like granulation tissue with lymphocytic infiltration. The final pathological diagnosis was actinomycosis of the minor salivary glands. There has been no recurrence for two years at present.

Discussion

Actinomycosis is a granulomatous inflammatory disease caused mainly by Actinomyces israelii, one of the resident oral flora and a gram-positive anaerobic rod, with mixed infection from other bacteria. It manifests in the form of a suppurative, granulomatous inflammation. Approximately 80% of Actinomyces infections occur in the mandible, and around 1% occur in the minor salivary gland, which is extremely rare.

Sometimes actinomycosis occurs at sites in the abdomen, chest and brain. It is presumed that infection of the respiratory organ is caused by misswallowing of oral bacteria and that infection of the digestive apparatus is caused by swallowing oral bacteria. In the case of mandibular infection, multiple abscesses, board-like induration and tris-
mus are typical signs of actinomycosis. When it extends deeper into the bone, the infected area develops into osteomyelitis with sequestrum, and sometimes becomes very serious. As for reporting of actinomycosis with minor salivary gland infection, only a few cases of the lesion have been reported in the buccal and labial mucosa of minor salivary glands, and so on. In this case, which occurred in the buccal mucosa, the only symptom was a sense of incongruity due to mass formation. It is hard to distinguish actinomycosis from general purulent inflammation for use of antibiotic therapy and monitoring disease progression. The final diagnosis of actinomycosis is done by identifying Actinomyces bacteria through culture examination or histopathological examination. The causative organism is arduous to cultivate, which makes diagnosis difficult even when the condition is strongly suspected. In this case, we did not cultivate the organism, but we could make the diagnosis of actinomycosis by histopathological examination including gram and Grocott staining. Furthermore, we carried out Ziehl-Neelsen staining to distinguish Actinomyces from similar filamentous bacteria of the Nocardia genus. Actinomyces are not acid-fast and do not stain with Ziehl-Neelsen staining but Nocardia do stain well. In this case, Ziehl-Neelsen staining was negative.

Actinomyces israelii usually exists as part of the normal flora of the oral cavity especially in caries cavities, periodontal pockets and tonsilla. It has been reported that the cause of infection is surgical intervention or traumatic invasion such as pericoronitis, tooth extraction, denture bite trauma of the buccal mucosa, and foreign body invasion. However, there have been a few cases where the focus of infection could not be specified. It has been reported that dental infection is the most common cause of actinomycosis (60%)\textsuperscript{8–10}. In this case, the patient complained of maladaptation of his dentures, so we suspect the cause of infection was denture bite trauma of the buccal mucosa. Histopathological findings revealed basophilic amorphous masses of Actinomyces and granulation tissue, and a fibrous capsule with lymphocytic infiltration surrounding the dilated excretory duct with squamous metaplasia, indicating that the inflammatory reaction arose from denture bite trauma of the buccal mucosa.

Treatment consists of surgical drainage of the infection and antibiotic therapy. Penicillin has proved to be the drug of choice. There have been reports that tetracyclines and carbapenems are effective for the treatment of actinomycosis\textsuperscript{11}. In this case, we administered cefem-type antibiotics after surgery. The region was not infected after surgery and the clinical course was good.

It is important to distinguish actinomycosis arising limited to the soft tissue (sometimes aris-
ing as an elastic, hard and unclear borderline mass) from a tumor. Upon clinical diagnosis, MRI and US have been reported to gain higher availability, but there are few reports about imaging of actinomycosis. At the time of clinical diagnosis, a comprehensive judgment must be made using clinical signs, imaging findings and blood examination data. In this case, we just excised surgically after the final clinical diagnosis of infection of minor salivary gland was made because typical clinical symptoms of actinomycosis were insufficient for making the diagnosis. In such a case as this of actinomycosis arising limited to the soft tissue, the lesion lacks blood flow because of fiber formation, so it has been reported that administration of antibiotics should normally be prolonged. On the other hand, it was reported that surgical therapy, such as excision or incision and drainage, is effective for healing. Only surgical therapy for the lesion was performed in this case: there has been no recurrence and the prognosis after the operation is good.

In conclusion, we experienced a very rare case of actinomycosis arising in the minor salivary gland in the buccal region. Histopathological examinations such as gram, Zeihl-Neelsen, and Grocott staining were very useful for making the final diagnosis.

References