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Case report

Actinomycosis manifested in the buccal mucosa estimated with scraping cytology: A case report

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ABSTRACT

Actinomycosis, a rare chronic granulomatous bacterial infection induced by *Actinomyces* species, colonizes the mouth, colon, and vagina. Although *Actinomyces* spp. are well-known oral commensals, actinomycosis is a rare oral infection. It is difficult to detect *Actinomyces* in biopsies, using histopathologic methods, or through examination of bacterial cultures; therefore, providing an accurate diagnosis is problematic. Recently, oral brush scraping cytology has been performed for the diagnosis of premalignant and malignant lesions, but it is difficult to determine the efficacy of this method based on the published reports, and whether it is a suitable tool for detecting *Actinomyces* infections in the general public. Here, we report a rare case of actinomycosis of the buccal mucosa in an 8-year-old boy that was successfully diagnosed by scraping cytology. He exhibited pain and swelling in the left buccal, and was admitted to our hospital for emergency care. Clinical examination revealed pyrexia, tachycardia, trismus, ingestion disorder, lethargy, and ulcer as well as having an indurated mass in the left buccal mucosa accompanied by strong pain. Following a clinical diagnosis of acute phlegmon of the left buccal mucosa, antibiotic and fluid therapy was performed. Because of detection of *Actinomyces* colonies at the ulcer surface using scraping cytology, we continued to administer long-term antibiotic therapy with penicillin for 2 months. The patient's general condition was improved, and this was confirmed by laboratory results, and the trismus and painful nodular mass were successfully cured. The patient has shown no subsequent evidence of recurrence for 7 months of following treatment.

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1. Introduction

Actinomycosis is a chronic, suppurative, and granulomatous infectious disease. In humans, the disease is caused mainly by *Actinomyces israelii* that is a species of Gram-positive, rod-shaped bacteria within the genus *Actinomyces* [1,2]. Known to live commensally within the human oral cavity, *A. israelii* is an opportunistic pathogen and a cause of cervicofacial actinomycosis as well as

chest or abdominal infections. The main clinical symptoms of oral actinomycosis are painless or painful masses with board-like induration, the formation of multiple small abscesses, and trismus [1–3]. Moreover, positive results on bacterial culture examination, or histopathological detection of *Actinomyces* colonization in lesions or pus, is a necessary diagnostic criterion for this disease. Although common in the past, the presentation of these typical symptoms has become increasingly rare with the development and wider use of antibiotics; therefore, it is often difficult to accurately diagnose this disease. Scraping cytology is a type of exfoliative cytology, in which a brush is used to remove cells from the surface of lesions that can be observed in a smear preparation. This examination is superior to previous diagnostic methods, both in terms of the relative convenience for clinicians in obtaining samples, and in the reduction of stress on patients as it is a comparatively non-invasive procedure. Similar tests have been applied to gynecological examinations to screen for various conditions, and to diagnose many

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Fig. 1. Image taken during the patient's first visit to our department, showing an ulcer, painful indurated mass, redness, and swelling of the left buccal mucosa.

kinds of diseases early and easily. Although these results demonstrate the efficacy of this technique when applied to mucosa in other organs, at present there are relatively few medical facilities that employ scraping cytology for oral cavity examinations, therefore resulting a few clinical reports of the use of this method for diagnosing premalignant and malignant oral lesions [4–6]. Here, we report the successful diagnosis of a rare case of actinomycosis of the buccal mucosa in an 8-year-old boy, through the application of scraping cytology.

2. Case report

An 8-year-old male patient was admitted to Department of Oral and Maxillofacial, Surgery Division of Dentistry, Tohoku University Hospital in September 2015 for emergency treatment, on complaint of pain in the left buccal mucosa and experiencing the opening difficulty of his mouth. The patient had no other recent history of infection related to injury, of related conditions, or of having taken prescribed medication. Physical examination revealed signs of an infection disorder, including cervical lymph node swelling, ulcer, a painful indurated mass, and redness and swelling of the left buccal mucosa, but revealed no fever (body temperature at time of examination, 37.3 °C), drainage of pus from the buccal mucosa, or any abnormal blood test results, including inflammatory markers, at that time (Fig. 1). Only normal flora was detected in bacterial culture taken from a swab of the ulcer surface. A panoramic radiograph showed no abnormal findings in the teeth or jaw. He reported an incident in which he had accidentally bitten his left buccal mucosa. The clinical diagnosis was a buccal ulcer, related to the bite. The patient was treated with cefditoren pivoxil (3 × 0.1 g/day), and was then required to irrigate the affected area with normal saline until discharge. Ten days after the first oral administration of antibiotic, most of these symptoms had improved.

In October 2015, 14 days after the first visit, the patient presented again with pain and swelling in the left buccal mucosa, and was readmitted to our hospital for emergency treatment. Clinical examination revealed pyrexia (38.5 °C), tachycardia (heart rate, 106 beats/min), trismus (maximum 5 mm), an ingestion disorder, lethargy, and an increase in the size of the ulcer and indurated mass in the left buccal mucosa, with severe associated pain (Figs. 2 and 3).

Laboratory findings indicated the presence of acute systemic inflammation (WBC 18,500/mm³, C-reactive protein 1.0 mg/dL). The patient was admitted to a pediatric ward in our hospital, following the diagnosis of acute phlegmon of the left buccal mucosa, resulting from recurrence of infection of the original lesion. The infection of the buccal mucosa was treated with irrigation, followed by intravenous administration of the antibiotic cefmetazole (3 × 0.5 g/day). During the acute phase, bacterial culture exami-

nation and scraping cytology of the ulcer were performed. The scraping cytology method was performed according to the guidelines of the Japanese Association of Clinical Cytology in 2016. The patient's mouth was washed before the procedure, and smears were taken from buccal mucosa under adequate illumination, by scraping with a sterilized commercially available nylon toothbrush. Using moderate pressure, the brush was repeatedly brushed in one direction over the entire lesion until pinpoint bleeding was obtained, and cells were extracted from the lamina propria or fat tissue layer. The material from the brush was spread onto the middle third of two clean, dry glass slides. The smears were fixed immediately with 95% isopropyl alcohol for staining using the Papanicolaou method. Significant bacterial colonization was not detected at the ulcer surface in bacterial culture examination. However, on cytological examination of the brush cytology oral smears, multiple colonies of *Actinomyces* were seen, in conjunction with other rod-shaped bacteria and epithelial tissue, and an inflammatory background. *Actinomyces* strangles with abundant mycelia radiating outwards are visible, accompanying neutrophilic inflammation (Fig. 3). Based on the guidelines of the Japanese Association of Clinical Cytology in 2016 with a cytotechnologist and two cytopathologists, the cytological results in accordance with the Bethesda System were negative for an intraepithelial lesion or malignancy. MR image acquired on day 3 showed an oval lesion of the left buccal mucosa, and indicated local granulomatous inflammation and the expanse of the abscess (Fig. 4).

Considering these findings, we diagnosed the patient with actinomycosis of the buccal mucosa, and treated this infection by irrigation with the long-term antibiotic amoxicillin (3 × 0.5 g/day). The patient showed an improvement in general condition, with an increase in appetite and recovery from swelling and trismus. For this reason, the patient was discharged on day 5 (Fig. 5). Long term antibiotic therapy with amoxicillin was continued for 2 months, with continued monitoring of the patient's condition after discharge. The ulcer and indurated mass were eliminated within 1 month after discharge, and the patient showed no evidence of recurrence within the 7-month follow-up period following the end of treatment with amoxicillin (Fig. 6).

3. Discussion

Actinomycosis of the cervicofacial area is mainly attributed to infection with *A. israelii*. *Actinomyces* are normal commensal bacteria of the oral cavity, and colonize the mucosa, dental plaque, tartar, and tonsil fossa. These bacteria are considered to have low pathogenicity, and actinomycosis is generally developed as a result of concomitant or opportunistic infection [1–3]. This disease frequently develops in the maxilla and mandible bones [3]. Conversely, this infection is rarely confined only to soft tissue, as we presented in this case. It is generally believed that the contributing factors to the development of actinomycosis on this occasion were remaining suture threads from the initial treatment for buccal ulcer and a secondary infection resulting from oral surgery and bites [7–10].

For a definitive diagnosis of this disease, the existence of an *Actinomyces* colony, or detection of *Actinomyces* by bacterial culture, is required in addition to clinical symptoms, including board-like induration, formation of multiple small abscesses, and trismus [1,2]. However, the detection of *Actinomyces* in bacterial cultures is difficult, because of contamination by the normal bacterial flora from the oral cavity and the added complication of producing a suitable anaerobic culture [3,11]. In addition, it is thought that the growth of these bacteria is effected by many antibiotics, and that colonies can become latent as result of initial antibiotic therapy, therefore making their detection more difficult [3,12]. Many cases

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