



Actinomycosis of the parotid gland: case series

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Background: Actinomycosis of the parotid gland is very rare. We sought to examine the clinical features, treatment methods, and treatment results of actinomycosis of the parotid gland in our hospital.

Methods: We retrospectively enrolled 5 patients with histopathologically identified actinomycosis of the parotid gland from January 2010 to May 2020.

Results: This study included 3 male and 2 female subjects. All patients had a common complaint of the mass in the parotid gland. Skin necrosis was observed in one patient. However, skin necrosis and fistula track occurred in the other 2 patients after fine-needle aspiration cytology (FNAC). Based on the result of FNAC two cases of inflammation and abscess were identified. Four patients with suspicion of parotid tumors before surgery underwent parotidectomy, and one patient with consideration of parotid abscess underwent incision and drainage. After surgery, intravenous administration of antibiotics was performed for an average of 6 days and oral antibiotics were prescribed for about 2 months for patients with actinomycosis of the parotid gland.

Conclusions: In case of skin necrosis of the parotid lesion without evidence of a malignant tumor, or if FNAC does not reveal the presence of cancerous cells, and fistula tract or skin necrosis occurs after the FNAC, then actinomycosis of the parotid gland should be suspected. If the actinomycosis of the parotid gland was localized to the parotid gland and removed surgically, it would be sufficient to treat the patients with antibiotics for about 2 months while observing the clinical response.

Keywords: Parotid gland; actinomycosis; antibiotics; surgical procedures; fine-needle biopsy

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Introduction

Actinomycosis is a rare granulomatous lesion characterized by the formation abscess, fibrosis of tissues, and draining sinuses, usually caused by *Actinomyces israelii* (1-6). Actinomycosis is classified according to the anatomical site infected as cervicofacial (55% of cases, most common), thoracic, abdominopelvic, and central nervous system, of which cervicofacial is the most common type (1,2,4,5). However, parotid actinomycosis is very rare, so only a few cases have been reported since Leonarmant first reported primary actinomycosis of parotid gland in 1913

(2-9). Actinomycosis requires a differential diagnosis from parotid lesions, including benign tumor, carcinoma, tuberculosis, cellulitis, fungal infections, and pyogenic abscess (2-4,6-9). The main treatment for actinomycosis is antibiotic therapy, but the exact duration of treatment has not been established (1-11).

Therefore, we sought to examine the clinical features, treatment methods, and treatment results of actinomycosis of the parotid gland in our hospital.

We present the following article in accordance with the AME Case Series Checklist (available at <http://dx.doi.org/10.21037/apm-21-755>).

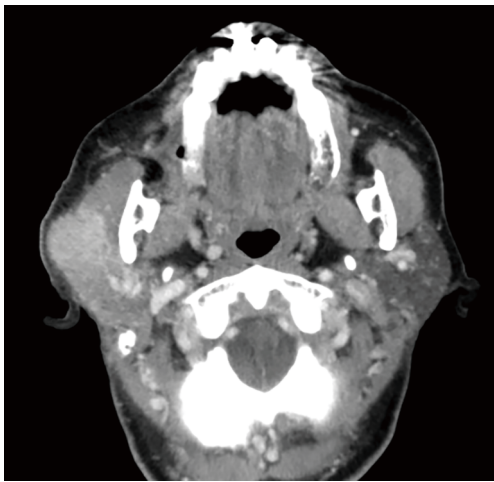


Figure 1 A 72-year-old female patient with actinomycosis of the parotid gland. Neck CT scan shows a 3 cm × 2.5 cm × 3.3 cm heterogeneously enhancing mass with perilesional infiltration in the right parotid gland.

Methods

All patient samples were treated in accordance with the Declaration of Helsinki (as revised in 2013), and the study was approved by the independent ethics committees at Chonnam National University Hwasun Hospital (CNUHH-2020-141), written informed consent was obtained from the patient. A review of 968 patients who underwent parotid surgery at this hospital for about 11 years from 2010 to 2020 confirmed 5 patients (5/968, 0.005%) diagnosed with actinomycosis in the final biopsy after surgery. The patients' clinical data were reviewed for age, sex, underlying diseases, a dental problem, trauma, previous infection, location, symptoms, duration of symptoms, fine-needle aspiration cytology (FNAC), diagnostic method, skin status, preoperative diagnosis, surgical procedures, microbiological culture, postoperative treatment, complications, and recurrence.

All patients underwent computed tomography (CT) before surgery for the assessment of the extent of the lesions and to aid in treatment planning (*Figure 1*). Preoperative skin status was divided into intact and necrosis. All patients underwent surgery, and the histopathological examination confirmed actinomycosis.

Statistical analysis

The results were presented as median and range as well

as mean ± SD. The quantitative data were analyzed with Student's *t*-test using Statistical Package for the Social Sciences version 20.0 (IBM Corporation, Armonk, NY, USA). $P < 0.05$ was considered a statistically significant value.

Results

The clinical data of the five patients with actinomycosis of the parotid gland are summarized in *Table 1*. This study included 3 male (60.0%) and 2 female (40.0%) subjects. The age of patients ranged between 21 and 74 years, with a mean of 56.4 ± 21.4 years. Only one patient had hypertension as the underlying disease, and the other four had no previous medical problems. The patients did not undergo any dental treatment recently and had no complaints of dental abnormalities such as discomfort in their teeth. One patient underwent a facelift procedure at the same site a year before the onset of the disease. Two patients had swelling or pain in the same area 3 months back.

All patients complained of the parotid mass as the main symptom. The duration of the symptom was 1.4 ± 1.5 months (range, 0.3 to 4 months). Among the five lesions, four (80.0%) were in the right parotid gland and one (20.0%) was in the left gland. The size of the lesion was 2.8 ± 1.2 cm (range, 1.2 to 4.4 cm). The preoperative skin status was mainly intact ($n=4$). Necrosis was observed in one patient. However, skin necrosis occurred in 2 of 4 FNAC patients. Based on the result of FNAC two cases of inflammation and abscess were identified

Four patients with suspected parotid tumor prior to surgery underwent superficial parotidectomy with facial nerve preservation (*Figure 2*), and one patient with probable presence of parotid abscess underwent incision and drainage. There was no major surgical complications including, facial nerve palsy. The culture test in 3 patients revealed the absence of actinomycosis. After surgery, mainly cephalosporin was administered intravenously for an average of 6 days (range, 5 to 9 days). In one patient, initially cephalosporin was administered and later replaced with penicillin after the diagnosis of actinomycosis based on the recommendation of the Department of Infectious Diseases. Subsequently, oral penicillin was prescribed for an average of 57.8 days (range, 28 to 105 days). The duration or dose of antibiotic treatment was determined in consultation with the Department of Infectious Diseases after checking the severity of the disease and the patient's symptoms.

The mean follow-up period after surgery was 40.0 ± 31.2 months, with a range from 1 to 76 months. One

Table 1 Clinical data of five patients with actinomycosis of the parotid gland

Age (years/sex)	PRI	Symptom (Du, Mo)	Site	Size (cm)	Skin	FNAC	Preop. Ds.	Surgery	IV anti (Du, D)	Oral anti (Du, D)	F/U	Recur
57/M		Mass [1]	R	3	Intact [†]	Inflammation	Tumor	Excision	Cepha [5]	PC [28]	16	
57/F*	Y	Mass [0.3]	L	2.3	Intact	Abscess	Tumor	Excision	Cepha [5]	PC [88]	76	Y
21/M		Mass [1]	R	4.4	Necrosis		Infection	I & D	Cepha [8]	PC [35]	62	
74/M	Y	Mass [0.5]	R	1.2	Intact	Inflammation	Tumor	Excision	Cepha [3]	PC [105]	45	
73/F [†]		Mass [4]	R	3.3	Intact [†]	Abscess	Tumor	Excision	Cepha [4] + PC [5]	PC [33]	1	

*The patient underwent a facelift procedure on the same site; [†]The patient had hypertension in past medical history; [‡]In these two patients, the skin at the lesion site was normal at first, but necrosis developed after FNAC. PRI, previous infection; Du, duration; Mo, months; cm, centimeter; FNAC, fine-needle aspiration cytology; Preop. Ds, preoperative diagnosis; IV anti, intravenous antibiotics; D, days; Oral anti, oral antibiotics; F/U, follow-up; M, male; F, female; Y, yes; R, right; L, left; I & D, incision and drainage; Cepha, cephalosporin; PC, penicillin.



Figure 2 A 72-year-old female patient with actinomycosis of the parotid gland. The intraoperative photograph demonstrates that parotid actinomycosis has been removed cleanly.

patient had relapsed after two years of completion of the first treatment and demonstrated improvement with antibiotic treatment for two weeks. Since then, there have been no cases of relapse.

Discussion

Five patients with complaints of actinomycosis of the parotid gland for the past 10 years were mainly diagnosed with parotid tumors before surgery. Consequently, the characteristics of the actinomycosis of the parotid gland are discussed in the present study.

Actinomycosis is usually caused by *Actinomyces israelii*, which is gram-positive, anaerobic bacteria, and normal inhabitants of the human oral cavity (1,2,4,5,10). Actinomycosis is not normally pathogenic, and predisposing conditions include dental caries and extractions, infections, facial trauma, and local tissue damage by surgery (1,6,7,11). There were no medical or dental problems among our patients. However, one person who underwent a facelift procedure is believed to have developed actinomycosis of the parotid gland.

Actinomycosis of the parotid gland appears as a chronic, and slow-growing swelling (1,7,9,10). With the progression of the disease, the development of the fistula tract and discharge occurs (7,9). All the 5 patients complained of swelling and presence of a mass in the parotid gland, and one of them had already developed a fistula at the time of visit. Skin necrosis and fistula tract occurred in the other 2 patients after FNAC (Figure 3). We thought that actinomycosis had progressed to the skin through the FNAC needle passage.



Figure 3 A 72-year-old female patient with actinomycosis of the parotid gland. Surgical specimen with skin necrosis and fistula tract after fine-needle aspiration cytology (FNAC).

It is difficult to differentiate actinomycosis from parotid tumors based on imaging tests including CT, ultrasound (US), magnetic resonance imaging (MRI), and positron emission tomography (PET) (1,3,5,6,8-11). In the present study 4 patients were diagnosed with parotid tumors and 1 patient with parotid abscess before surgery. The definitive diagnosis of actinomycosis of the parotid gland is made by microbiologic or histopathologic findings (1-11). However, the culture of *Actinomyces* is difficult due to the anaerobic nature of the organism (4,5,10). The diagnosis of *Actinomyces* in all 3 patients who underwent culture could not be achieved. FNAC or surgical tissue biopsy is essential for accurate diagnosis (1-11). FNAC is known as an easy, safe, and rapid procedure (1,2). However, all four patients who underwent FNAC exhibited failure in the diagnosis of actinomycosis. Consequently, in the absence of clinical and radiological differentiation, and lack of FNAC based diagnosis of actinomycosis from the parotid tumor, it is necessary to perform surgery based on the location of parotid tumors for accurate diagnosis and treatment (3,5,9).

Treatment for actinomycosis of the parotid gland involves antibiotic therapy and surgery (1-11). Penicillin is the drug of choice, but doxycycline, erythromycin, and cephalosporin may be used (1,4,5,7,9,10). However, recommended treatment regimens vary from two weeks to several months (1,4,5,10). Recently, the duration or dose of antibiotics therapy has been determined according to the severity of the disease and the response to treatment (1,10,11). We determined the duration or dose of antibiotic treatment in consultation with the Department of Infectious Medicine

after checking the severity of the disease and the patient's symptoms. In the present study, 6 days of intravenous antibiotics administration (on an average) and prescription of oral antibiotics for about 2 months were chosen as the preferred modes of treatment. Surgery for actinomycosis of the parotid glands is mandatory, in cases of extensive abscess, necrotic tissue, fistula tract, or absence of response to antibiotic therapy, and lack of differentiation from parotid tumors (1,4,5,9-11). Four patients with suspicion of parotid tumors underwent superficial parotidectomy before surgery with a safety margin like parotid tumors. One patient with plausible parotid abscess underwent incision and drainage.

The prognosis for actinomycosis of the parotid gland is very good and exhibits improvement in most of the cases (1). All our patients demonstrated improvement with surgery and antibiotic therapy. However, based on the case of one patient with relapse two years after the first treatment, it is proposed that the clinical response should be closely monitored to prevent local recurrence (10).

The limitation of this study is that it is a retrospective study and the number of cases is small.

Conclusions

Actinomycosis of the parotid gland is very rare, and it is difficult to distinguish it from the parotid tumors based on clinical features or radiologic examinations. In case of skin necrosis of the parotid lesion without evidence of a malignant tumor, or if cancerous cells are not identified in FNAC and fistula tract or skin necrosis occurs after the FNAC, actinomycosis of the parotid gland should be suspected. If the actinomycosis of the parotid gland is localized to the parotid gland and surgically removed, it would be sufficient to treat the patients with antibiotics for about 2 months while observing the clinical response.

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Footnote

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All patient samples were treated in accordance with the Declaration of Helsinki (as revised in 2013), and the study was approved by the independent ethics committees at Chonnam National University Hwasun Hospital (CNUHH-2020-141). And written informed consent was obtained from the patient.

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