

Case Report**Clinical and Histopathologic Investigation of Periapical Actinomycosis with Cutaneous Lesion: a Case Report**Davoud Jamshidi ^a, Fariborz Moazami ^b, Fereshteh Sobhnamayan ^b, Ali Taheri ^c^a Dept. of Endodontics, School of Dentistry, Qazvin University of Medical Sciences, Qazvin, Iran.^b Dept. of Endodontics, School of Dentistry, Shiraz University of Medical Sciences, Shiraz, Iran.^c Dept. of Pathology, School of Dentistry, Kerman University of Medical Sciences, Kerman, Iran.**KEY WORDS**Actinomycosis
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Accepted January 2015;**ABSTRACT**

Management of an extra-radicular infection is a challenging procedure that requires surgical intervention. This report describes a patient with discharging cutaneous lesion that required apical surgery. A 40-year-old woman was referred to the Department of Endodontics, Shiraz Dental School with chief complaint of a cutaneous sinus tract. She had been treated by a dermatologist and an otolaryngologist. The patient had also received orthograde root canal treatment of tooth #16. Yet, the lesion was still discharging and the patient was scheduled for surgery. Histopathologic analysis of the lesion showed actinomycosis infection. A 36-month follow-up revealed clinical and radiographic healing.

Corresponding Author: Jamshidi D., Dept. of Endodontics, School of Dentistry, Qazvin University of Medical Science, Bahonar Blvd., Qazvin, Iran. Tel: +98-28-33353061-2
Fax: +98-28-33353066 Email: davoud.jamshidi@gmail.com, djamshidi@qums.ac.ir

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Introduction

Apical periodontitis is an inflammatory disease which is formed in response to intraradicular infection [1] and usually constitutes a barrier against the spread of infection to the bone. [2]

Microorganisms are not usually present in apical periodontitis lesions, [3] but in an extra-radicular infection, bacteria can invade the inflamed periradicular tissues. [4] In such conditions, extra-radicular bacteria have been found in biofilms adhering to the apical root surface [5] or located within the body of inflammatory lesion, usually forming cohesive actinomycotic colonies. [6]

The extra-radicular infection can be dependent on or independent of the intraradicular infection. For example, acute apical abscess is mostly dependent on intraradicular infection [4] and periapical actinomycosis is an independent extra-radicular infection [2] caused by anaerobic or facultative gram-positive, non-spore-forming, filamentous rod bacteria. These bacteria are not virulent and produce chronic, slow-developing, opportunistic granulomatous infections.

[7] Strains of actinomyces have been associated with cases of failed endodontic therapy. [8]

This paper reports a case of extra-radicular actinomycosis that caused failure of endodontic treatment, and thus made apical surgery necessary.

Case Report

A 40-year-old Afghan female patient was referred to the Endodontic Department of Shiraz Dental School by an endodontist. She was complaining about a persistent cutaneous lesion on the right cheek of her face persistent for more than a year.

She had been treated by a dermatologist and an otolaryngologist but the lesion was not cured. Then, she was referred to an endodontist. Root canal therapy of her tooth #16 was completed in two sessions, calcium hydroxide slurry was placed for a week, and the tooth was restored with amalgam. After a month, the patient returned with persistent sinus tract discharge. In clinical examination, the cutaneous lesion expressed a purulent discharge (Figure 1a). The tooth had normal periodontal probing and mobility. The alveolar mucosa

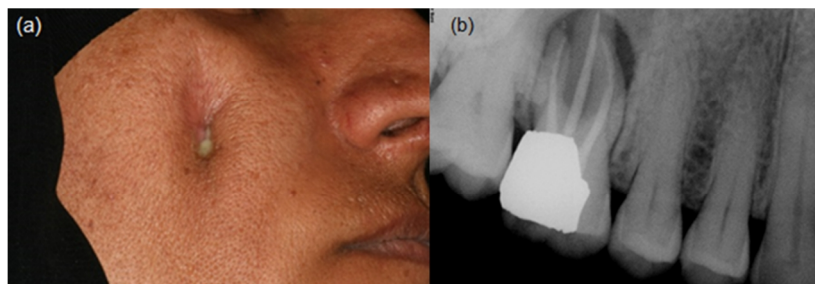


Figure 1: Radiograph and photograph of the patient in the examination session: **a:** Lateral view showing discharge. **b:** A periapical radiograph showing tooth #16 with radiolucency and complete root canal filling.

adjacent to the tooth was not tender to palpation. The patient had a non-contributory medical history.

Radiographic examination revealed periradicular radiolucencies associated with teeth #16 (Figure 1b).

The quality of the root canal filling was good radiographically. Periradicular surgery was, therefore, arranged. The patient was informed of the treatment plan and her written consent was obtained before treatment.

Chlorhexidine gluconate (0.2%) was used as a mouth rinse to reduce the number of surface microorganisms in the surgical field. After local anesthesia (2% lidocaine and 1:80000 epinephrine; Darupakhsh, Tehran, Iran) triangular full-thickness mucoperiosteal flap was reflected from teeth #14 to #17. Periapical pathological lesion was noted at all the three apices of this tooth. The granulation tissue was excised and sent for histologic evaluation. Osteotomy and apicoectomy of mesiobuccal, distobuccal, and palatal roots of tooth #16 were performed and retropreparations were made using an ultrasonic tip (E3ID; NSK, Japan). The root-end cavity was filled with mineral trioxide aggregate (ProRoot MTA; Dentsply, Tulsa dental, OK, USA). All procedures were performed at high magnification (OPMI Pico Dental Microscope; Zeiss, Oberkochen,

Germany).

The flap was sutured with 3-0 silk sutures (Ethicon; PA, USA). The patient received postoperative instructions. Postoperative analgesic (Ibuprofen 400 mg four times a day) and mouth rinse (Chlorhexidine 0.2%, 15 ml twice a day) were prescribed. The sutures were removed after four days. The patient reported a slight pain and no sign of discharge were observed. Histopathologic findings confirmed actinomycosis infection (Figure 2a, b).

In the 6-month recall session, the patient had no discomfort, so she was referred to a plastic surgeon (Figure 3a, b). In the 36-month follow up, the patient was asymptomatic and revealed no sign of discharge. Radiographic examination also showed periapical healing (Figure 3c). In addition to these findings, cutaneous deficiency was corrected through plastic surgery (Figure 3d).

Discussion

Microorganisms play an important role in the etiology and development of periradicular inflammatory responses. [1] Although nonsurgical endodontic treatment has been successful in the majority of cases, some of these inflammatory lesions do not heal by these treat-

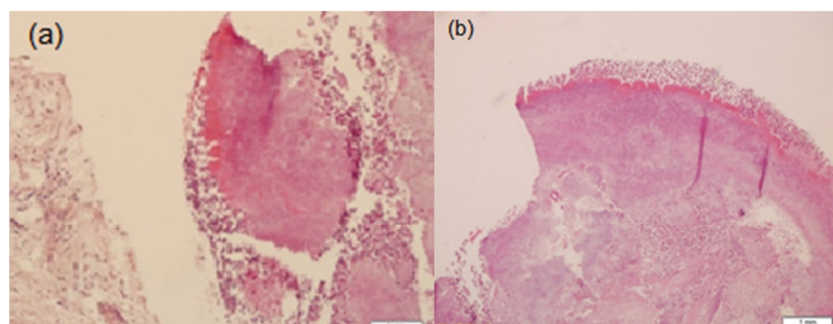


Figure 2: Micrograph of the lesion. **a:** The section shows colony of actinomycotic organisms surrounded by polymorphonuclear leukocytes with a part of connective tissue (×100). **b:** On higher magnification (×200), the section shows the rosette pattern of bacterial filaments. The bacterial colony has a central basophilic core and a peripheral eosinophilic portion which is surrounded by neutrophils.

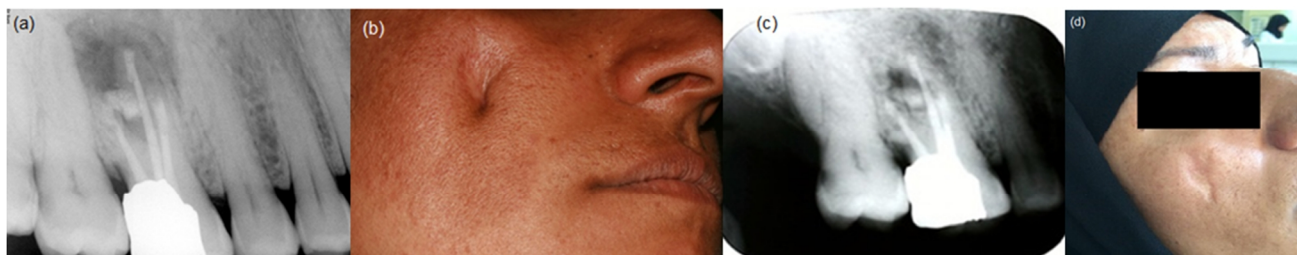


Figure 3a: A radiograph after 6 months. **b:** A photograph in the 6-month recall. **c:** A radiograph after 36 months. **d:** A photograph after plastic surgery in the 36-month follow-up.

ents. [9] The most common factor associated with lack of healing is the presence of microorganisms. [10] This failure is commonly related to the presence of residual bacteria (persistent infection) or reinfection of a previously disinfected root canal environment (secondary infection). [11] Actinomyces species have been reported in persistent and secondary intraradicular infections. [10]

The actinomycotic organisms are able to establish extra-radicular, so they can perpetuate even after proper root canal treatment. [8] Because they are inaccessible to endodontic disinfection procedures, these microorganisms may be a factor in the failure of root canal therapy.

If a sinus tract does not close after appropriate removal of the primary etiology, actinomycosis is the most common cause of the lesion. [12] A case report showed that actinomycosis infection may remain even after extraction of infected tooth. [13]

Post-treatment apical pathologies are managed by nonsurgical retreatment, apical surgery, or extraction and implant placement. [10] Periradicular surgery is often the last resort to maintain a root filled tooth with a persistent periapical lesion. [14]

Because of sufficient root canal treatment, it seemed that nonsurgical root canal treatment would not yield a better result and consequently, surgical treatment was indicated in this case. [15]

Periapical actinomycosis have been reported as a cause of endodontic failure; [8] however, they should not be considered a common cause of endodontic failure because of their low-prevalence infections. [16]

The incidence of this infection was reported to be 1.8% by Hirshberg *et al.* [16] and 4.4% by Nair and Schroeder. [17] Although Xia *et al.* [18] used PCR (polymerase chain reaction) and demonstrated actinomyces species to be often present in endodontic failure

and their frequency to be higher than previously believed, most publications on this pathologic entity are in the form of case reports. [16, 19-20]

An odontogenic cutaneous sinus tract is a pathologic channel that originates in the oral cavity but exits at the cutaneous surface of the face or neck. [21]

Nearly 80% of the reported cases of odontogenic cutaneous sinus tracts have been associated with mandibular teeth [21-25] and cutaneous sinus tracts on the upper part of the cheek are most likely associated with the maxillary posterior teeth. [21, 23, 25]

Periapical actinomycosis is a main cause of cutaneous sinus tracts. [26] The differential diagnosis include furuncles, bacterial infections, carcinomas, osteomyelitis, pyogenic granulomas and congenital fistulas, [25] squamous cell carcinoma, thyroglossal duct cyst, branchial cleft cyst, and epidermal cyst. [27]

Due to the rarity of odontogenic cutaneous sinus tracts and the absence of dental symptoms in these patients, misdiagnosis is common. [21] Hence, the patients have multiple appointments with medical practitioners prior to a correct diagnosis.

To make the correct diagnosis, clinical and medical history, pulp and periradicular diagnostic testing, radiographic findings, tracing the sinus tract and sinus tract angiography are imperative. [25, 28]

Clinical and radiographic manifestations of actinomycosis infections are not usually distinguishable from apical periodontitis, and multiple sinus tracts are not a prerequisite for diagnosis of apical actinomycosis. [16, 19] Definitive diagnosis of this pathologic lesion is, therefore, established by histologic and microbial assessment of the surgical specimen. [20]

Strains of actinomyces are sensitive to sodium hypochlorite and calcium hydroxides, thus, prescription of antibiotics are not indicated. [29]

Although periapical surgery can resolve cutaneo-

us sinus tract in 1 to 2 weeks, [30] fixation of the tract with the underlying tissues can cause cutaneous retraction or dimpling. [22] Therefore, the residual umbilication of the skin can be revised by cosmetic surgery. [31]

Conclusion

This case highlights the importance of dental examination in diagnosis of a cutaneous sinus tract and consequently confirms that a decent cooperation between physicians and dentists is indispensable in better diagnosis and treatment of such challenging cases.

Conflict of Interest

None declared

References

- [1] Kakehashi S, Stanley HR, Fitzgerald RJ. The effects of surgical exposures of dental pulps in germ-free and conventional laboratory rats. *Oral Surg Oral Med Oral Pathol.* 1965; 20: 340–349.
- [2] Siqueira JF Jr. Endodontic infections: concepts, paradigms, and perspectives. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002; 94: 281-293.
- [3] Walton RE, Ardjmand K. Histological evaluation of the presence of bacteria in induced periapical lesions in monkeys. *J Endod.* 1992; 18: 216-227.
- [4] Sunde PT, Olsen I, Debelian GJ, Tronstad L. Microbiota of periapical lesions refractory to endodontic therapy. *J Endod.* 2002; 28: 304-310.
- [5] Tronstad L, Barnett F, Cervone F. Periapical bacterial plaque in teeth refractory to endodontic treatment. *Endod Dent Traumatol.* 1990; 6: 73-77.
- [6] Happonen RP. Periapical actinomycosis: a follow-up study of 16 surgically treated cases. *Endod Dent Traumatol.* 1986; 2: 205-209.
- [7] Rush JR, Sulte HR, Cohen DM, Makkawy H. Course of infection and case outcome in individuals diagnosed with microbial colonies morphologically consistent with *Actinomyces* species. *J Endod.* 2002; 28: 613-618.
- [8] Nair PN. On the causes of persistent apical periodontitis: a review. *Int Endod J.* 2006; 39: 249-281.
- [9] Sjogren U, Hagglund B, Sundqvist G, Wing K. Factors affecting the long-term results of endodontic treatment. *J Endod.* 1990; 16: 498-504.
- [10] Signoretti FG, Endo MS, Gomes BP, Montagner F, Tosello FB, Jacinto RC. Persistent extraradicular infection in root-filled asymptomatic human tooth: scanning electron microscopic analysis and microbial investigation after apical microsurgery. *J Endod.* 2011; 37: 1696-1700.
- [11] Siqueira JF Jr. Reaction of periradicular tissues to root canal treatment: benefits and drawbacks. *Endod Topics* 2005; 10: 123–147.
- [12] Braun RJ, Lehman J 3rd. A dermatologic lesion resulting from a mandibular molar with periradicular pathosis. *Oral Surg Oral Med Oral Pathol.* 1981; 52: 210-212.
- [13] Göçmen G, Varol A, Göker K, Basa S. Actinomycosis: report of a case with a persistent extraoral sinus tract. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2011; 112: 121-123.
- [14] von Arx T, Peñarrocha M, Jensen S. Prognostic factors in apical surgery with root-end filling: a meta-analysis. *J Endod.* 2010; 36: 957-973.
- [15] Gutmann JL, Harrison JW. Posterior endodontic surgery: anatomical considerations and clinical techniques. *Int Endod J.* 1985; 18: 8-34.
- [16] Hirshberg A, Tsesis I, Metzger Z, Kaplan I. Periapical actinomycosis: a clinicopathologic study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2003; 95: 614-620.
- [17] Ramachandran Nair PN, Schroeder HE. Periapical actinomycosis. *J Endod.* 1984; 10: 567-570.
- [18] Xia T, Baumgartner JC. Occurrence of *Actinomyces* in infections of endodontic origin. *J Endod.* 2003; 29: 549-552.
- [19] Sjögren U, Happonen RP, Kahnberg KE, Sundqvist G. Survival of *Arachnia propionica* in periapical tissue. *Int Endod J.* 1988; 21: 277-282.
- [20] Ricucci D, Siqueira JF Jr. Apical actinomycosis as a continuum of intraradicular and extraradicular infection: case report and critical review on its involvement with treatment failure. *J Endod.* 2008; 34: 1124-1129.
- [21] Gupta M, Das D, Kapur R, Sibal N. A clinical predicament--diagnosis and differential diagnosis of cutaneous facial sinus tracts of dental origin: a series of case reports. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2011; 112: e132-e136.
- [22] Tidwell E, Jenkins JD, Ellis CD, Hutson B, Cederberg RA. Cutaneous odontogenic sinus tract to the chin: a case report. *Int Endod J.* 1997; 30: 352-355.
- [23] Mittal N, Gupta P. Management of extra oral sinus cas-

- es: a clinical dilemma. *J Endod.* 2004; 30: 541-547.
- [24] Herd MK, Aldridge T, Colbert SD, Brennan PA. Odontogenic skin sinus: a commonly overlooked skin presentation. *J Plast Reconstr Aesthet Surg.* 2012; 65: 1729-1730.
- [25] Bai J, Ji AP, Huang MW. Submental cutaneous sinus tract of mandibular second molar origin. *Int Endod J.* 2014; 47: 1185-1191.
- [26] Chowdri NA, Sheikh S, Gagloo MA, Parray FQ, Sheikh MA, Khan FA. Clinicopathological profile and surgical results of nonhealing sinuses and fistulous tracts of the head and neck region. *J Oral Maxillofac Surg.* 2009; 67: 2332-2336.
- [27] Held JL, Yunakov MJ, Barber RJ, Grossman ME. Cutaneous sinus of dental origin: a diagnosis requiring clinical and radiologic correlation. *Cutis.* 1989; 43: 22-24.
- [28] Nakamura Y, Hirayama K, Hossain M, Matsumoto K. A case of an odontogenic cutaneous sinus tract. *Int Endod J.* 1999; 32: 328-331.
- [29] Barnard D, Davies J, Figdor D. Susceptibility of *Actinomyces israelii* to antibiotics, sodium hypochlorite and calcium hydroxide. *Int Endod J.* 1996; 29: 320-326.
- [30] Spear KL, Sheridan PJ, Perry HO. Sinus tracts to the chin and jaw of dental origin. *J Am Acad Dermatol.* 1983; 8: 486-492.
- [31] Güleç AT, Seçkin D, Bulut S, Sarfakoğlu E. Cutaneous sinus tract of dental origin. *Int J Dermatol.* 2001; 40: 650-652.