An unusual case of periapical actinomycosis

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ABSTRACT
Actinomycosis is a granulomatous infection of the cervicofacial, thoracic, abdominal and cerebral regions. Periapical actinomycosis is a rare form of cervicofacial actinomycosis, which is a persistent lesion that usually does not respond to routine endodontic treatment. The prevalence of this infection is indeed low and therefore is not a common cause of endodontic failure. We report a case of periapical actinomycosis in a 24 yr old patient who presented with a complaint of swelling and pain in the maxillary incisors. The accidental finding of sulphur granules during the endodontic surgery led to the provisional diagnosis of periapical actinomycosis. Upon biopsy, actinomycosis was diagnosed where classic colonies were demonstrated. Apicoectomy was performed and the patient was maintained on follow up. Satisfactory healing was achieved after 10 months. Early diagnosis of such lesions with actinomyces is important since it directly impacts the treatment plan.

Keywords: Actinomyces israelii, antibiotic therapy, endodontic treatment, periapical actinomycosis, periapical surgery

INTRODUCTION
Actinomycosis was first described in the early 19th century as a disease in cattle. In 1845, Langenbeck was probably the first to describe the disease in humans.[1] This disease has also been recognized in 1854 by Graefe and in 1875 by Cohn. The first published clinical description of the entity was provided by Lebert in 1857.[2] Actinomycosis is a chronic infectious disease caused by filamentous, anaerobic, nonacid-fast Gram-positive rods from the Actinomyces family. At least 13 species are recognized,[3] with the most documented species being Actinomyces israelii, followed by Propionibacterium propionicum, Actinomyces naeslundii, Actinomyces viscosus, and Actinomyces odontolyticus.[3] A. israelii is a human commensal, usually isolated from supragingival and subgingival plaque, tonsils, dentinal and root surface caries, periodontal pockets, and infected root canals. Actinomyces are found in 10.6%–17.2% of infected root canals,[4,5] which happens to be the primary portal of entry of these organisms into the periapical tissues. Since these filamentous bacteria resemble fungi, it was misnamed Actinomyces but is actually a true bacterium. It produces a uniquely indolent inflammatory infection that forms a densely fibrotic lesion and slowly spreads to contiguous tissues without regard for tissue planes.[6] Sinus tracts are also formed frequently.

The usual location of actinomycosis is the cervicofacial region in 60% of cases, followed by the abdominal in 20%, thoracic in 15%, and the cerebral in 5%.[7] In the cervicofacial region, the organism typically enters the tissue through an area of prior trauma, such as a soft-tissue injury, periodontal pocket, nonvital tooth, extraction socket, or infected tonsil. Cervicofacial actinomycosis is usually associated with poor oral hygiene, a history of invasive dental procedures, or oral
trauma.\(^2\) Periapical actinomycosis is a cervicofacial form of actinomycosis. Periapical actinomycosis is defined as a nonresolving periapical lesion associated with actinomycotic infection and has been suggested as a contributing factor in the perpetuation of periapical radiolucencies after endodontic treatment.\(^4,7\) It has been regarded in the literature as a mild form of cervicofacial actinomycosis with a persisting periapical lesion that usually does not respond to routine endodontic treatment. Data regarding the occurrence of periapical actinomycosis and its correlation with cervicofacial actinomycosis are scarce. A few investigators have reported the incidence of periapical actinomycosis to be about 2\(^%-4\%\).\(^8,9\)

Sakellariou\(^7\) reviewed 45 reported cases of periapical actinomycosis.\(^10\) Since 1996, the total number of cases has increased to 94, although the number of surveys is small to imply a much greater frequency than this number would indicate.\(^4\) This is supported by Sjogren et al., according to whom the periapical actinomycosis could be more prevalent than reported.\(^11\)

CASE REPORT

A 24-year-old male patient reported to the institution with a chief complaint of pain, swelling, and pus discharge in the maxillary incisor region for 2 years. He also complained of discolored maxillary left central incisor. The patient gave a history of trauma about 2 years ago. On clinical examination, the presence of a sinus tract with pus discharge in teeth #21 and 22 was seen. Tooth #21 was found to be discolored [Figure 1]. Pulpal response of the teeth #21 and 22 on electric and thermal pulp vitality testing was negative. A periapical radiograph showed a well-defined radiolucency measuring approximately about 2 cm in diameter involving 21 and 22 [Figure 2]. A gutta-percha point was used to trace the origin of the sinus to tooth #21 in radiograph. Further, endodontic treatment was carried out for 21 and 22. Calcium hydroxide was given as an intracanal medicament, and obturation was completed by cold lateral condensation technique.

Due to the size of the lesion and recurrence of swelling, surgical endodontic treatment with local anesthesia was performed. Vertical releasing incisions were given, and full-thickness flap was reflected. Perforated underlying bone was seen on reflection of flap due to the cortical bone destruction caused by the lesion. Yellowish-gray granules were seen, accompanied by exudation of pus. The bone cavity was slightly enlarged with the help of osteotomy bur. The lesion had caused considerable destruction of the bone resulting in the total absence of the palatal bone. The lesion was curetted. A 3-mm apical resection was then performed with mineral trioxide aggregate (MTA) being used as a root-end filling material. Sutures were placed and removed after a week. After the surgery, the patient was prescribed Ciplox TZ (ciprofloxacin + tinidazole) and Enzoflam (diclofenac, paracetamol, and serratiopeptidase) TDS for 5 days. Bleaching was performed on 21 with satisfactory results. The patient was maintained on follow-up. A 10-month postoperative radiograph showed a resolution in the size of periapical radiolucency suggestive of a healing process [Figure 3].

The curetted tissue along with the yellowish-gray granules was sent for histopathological examination. Examination of the biopsy specimen using hematoxylin and eosin stain showed the presence of granulation tissue with diffuse chronic inflammatory infiltrate of plasma cells, lymphocytes, and fibroblasts. Numerous basophilic, fine-branching filaments of actinomycotic colonies surrounded with mixed inflammatory cells as well as a few basophilic granules [Figure 4] were seen. Periodic acid–Schiff stain confirmed the presence of actinomycotic colonies.

DISCUSSION

A. israelii is an anaerobic commensal microorganism of the human oral cavity. They become pathogenic once the mucosal barrier is compromised; they gain access to the deeper oral tissues and initiate a prolonged chronic inflammatory reaction. These microflora work synergistically to destroy the vascularized aerobic tissue and create poorly oxygenated granulation tissue, which becomes an environment that supports the growth and multiplication of Actinomyces. It provokes a chronic productive and colliquative inflammatory reaction which produces granulomatous and supplicative lesions by bacterial proliferation. A hallmark of this disease is the tendency to spread without regard for anatomical barriers, including facial planes and lymphatic channels.

Periapical actinomycosis is believed to be a persisting periapical lesion and has been suggested as a predisposing factor in the perpetuation of periapical lesions after root canal treatment.\(^8\) Because of their facultative anaerobic metabolism, they are also frequently isolated from the apical segment of infected canals and participate in the development of periapical abscesses, either by direct advance of the root canal infection or by apical extrusion of infected debris during root canal instrumentation.\(^6\) Egress of these organisms and their by-products from the infected root canal is considered
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the major cause of periapical pathosis. Definitive diagnosis is usually based on histologic identification of actinomycotic colonies, culture of Actinomyces, or both. Actinomycotic colonies consist of club-shaped filaments arranged in a radiating rosette pattern. The colonies are surrounded by a collection of polymorphonuclear leukocytes (Sunburst Fashions). Under hematoxylin-eosin staining, the center of the colony stains basophilic and the periphery appears eosinophilic. Methenamine silver stains work well for this purpose.

It is reported that the outcome of patients with periapical actinomycosis is excellent; in most cases, curettage of the lesion combined with a short course of antibiotics is sufficient to induce healing without complications. According to Sakellarion, endodontic therapy alone fails in these cases and requires surgical curettage of the lesion too.[7] In certain cases, despite adequate endodontic treatment, periapical lesion may persist and about 10% of well-treated cases end in failure.[11] This is because, according to Byström et al., Actinomyces have been shown to inhibit normal periradicular healing following conventional root canal therapy due to their capacity to survive in the periapical tissues beyond the root canal system.[8] The actual frequency of Actinomyces-associated endodontic treatment failures remains unclear.

It is noteworthy here to mention the role of the intracanal irrigants and medicaments in eradicating the infection. Several investigators have studied the same with considerable success.[12,13] Almost all the studies collaborate the fact that both sodium hypochlorite and calcium hydroxide are highly effective in inhibiting the growth of A. israelii. In addition, calcium hydroxide had inherent antibiotic activity against Actinomyces species.[14]

The use of antibiotics for treating periapical actinomycosis has been a subject of debate for long. Actinomycosis is sensitive to antibiotics that are effective against Gram-positive organisms such as penicillin, sulphonamides, streptomycin, tetracyclines, erythromycin, and rifampicin, but a high dose of long-term penicillin is the treatment of choice.[15] Data regarding the use of fluoroquinolones are limited; there have been anecdotal reports of cure with these antibiotics.[16,17] On the other hand, in a study by Smith et al. (2005), most of the Actinomyces species tested appeared resistant to ciprofloxacin.[18] Some investigators have routinely recommended that the use of high
concentrations of antibiotics such as penicillin is necessary to first penetrate the areas of dense fibrosis and then to kill the microorganisms, whereas disagree with the long-term use of antibiotics since recent studies have shown that in the treatment of localized cervicofacial (periapical) actinomycosis, the use of long-term antibiotics may not be necessary, since it is found to be especially responsive to a brief course of antibiotic treatment. If short-term antibiotic treatment is attempted, the clinical and radiological responses should be closely monitored.

Even though infected root canals provide a primary path of entry for the Actinomyces organisms into the periapical tissues, periapical actinomycosis is considered exceedingly rare. This may be due to the omission of routine histological examination of every periapical lesion and also the clinical behavior of the disease. In the cervicofacial forms, the clinical presentation is obvious: swelling, induration, and draining sinuses. On the contrary, periapical actinomycosis presents a very atypical picture which can be often confused with the more frequent chronic periapical inflammation.

CONCLUSION

The most common reason for relapse is a persisting extraradicular infection of the periapical tissues, which results in failure of endodontic therapy. One of the causes for repeated temporary resolution, followed by relapse, may indicate actinomycotic infection. Long-term (6–8 weeks) antibiotic therapy is warranted in cases where the infection has spread to contiguous structures. Such cases must be investigated and treated accordingly.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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