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## Surgical Treatment of Periapical Actinomycosis of Endodontic Origin: A rare case report

### Traitement Chirurgical D'une Actinomyose Périapicale D'origine Endodontique : A Propos d'un Cas Clinique Rare

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#### Abstract

**Aim :** Actinomycosis is a chronic and slowly progressive granulomatous disease caused by the filamentous Gram-positive anaerobic bacteria Actinomyces. The aim of the present case was to describe a rare case of periapical actinomycosis (PA) occurring in an adult patient.

**Case presentation :** The patient, a 66-year old type 2 diabetic man consulted the dental department urgently with a warm, red swelling located on the chin and associated with tooth 43. Information regarding patient's medical history and treatment were recorded. Clinical and radiological examinations were performed as well. The retroalveolar radiography showed inadequate endodontic fillings and radiolucent periapical images on tooth 42 and 43 (PAI>3). The Diagnosis of circumscribed cellulitis of the chin related to tooth 43 was retained. The tooth was opened through the metal crown, allowing drainage of an abundant pus. After mechanical preparation and disinfection, Ca (OH)2 paste, was then placed into the root canals of tooth 42 and 43. The patient received antibiotic therapy: Clamoxyl 2g/day for 7 days. Root canal fillings were carried out on tooth 42 and 43, but the fistula related to tooth 43 persisted with an increase of the peri apical lesion size. Surgical excision of the lesion was performed under local anesthesia. Histological analysis of the specimen showed a typical granulomatous tissue infected by actinomycotic granules. The outcome was marked by the healing of the draining fistula, 2 weeks after the surgery and the regeneration of the periapical tissues at one year.

**Conclusion :** In symptomatic persistent periapical lesions, with a draining fistula and an inadequate root canal filling, peri apical actinomycosis is the most probable diagnosis that the dentist should evoke.

#### Key words

Periapical Actinomycosis, Endodontic Retreatment, Chirurgie, Histopathology, Outcome

#### Résumé

**But :** L'actinomyose est une maladie granulomateuse chronique à évolution progressive, causée par des Actinomyces, bactéries à Gram positif, anaérobies et filamenteuses. Le but de ce travail est de décrire un cas rare d'actinomyose périapicale (AP) survenant chez un patient adulte.

**Présentation du cas :** Le patient âgé de 66 ans, diabétique de type 2 a consulté en urgence pour une tuméfaction chaude, rouge, localisée au niveau du menton en rapport avec la 43. Les informations concernant les antécédents médicaux et thérapeutiques ont été enregistrées. Les examens cliniques et radiologiques ont été aussi effectués. La radiographie rétroalvéolaire a montré des obturations canalaires inadéquates et des images périapicales radioclares en rapport avec la 42 et la 43 (IPA>3). Le diagnostic d'une cellulite circonscrite du menton en rapport avec la 43 a été retenu. L'ouverture de la dent à travers la couronne métallique a permis le drainage d'un pus abondant. Une pâte de Ca(OH)2 a été placée dans les canaux de la 42 et la 43 après préparation mécanisée et désinfection. Le patient a reçu une antibiothérapie à base de Clamoxyl 2g/jour pour 7 jours. Les obturations canalaires ont été effectuées sur les deux dents, mais la fistule en regard de la 43 a persisté avec une augmentation de la taille de la lésion périapicale. L'excision chirurgicale de la lésion a été faite sous anesthésie locale. L'analyse histologique de l'échantillon a montré un tissu granulomateux typique infecté par des granules d'Actinomyces. L'évolution a été marquée par la guérison de la fistule, 2 semaines après la chirurgie et la régénération du tissu périapical après un an.

**Conclusion :** Dans les lésions périapicales symptomatiques en présence d'une fistule et d'un traitement canalair inadéquat, le diagnostic le plus probable à évoquer par le médecin dentiste est l'actinomyose périapicale.

#### Mots clés

Actinomyose Périapicale, Retraitement Endodontique, Chirurgie, Histopathologie, Pronostic

## INTRODUCTION

Actinomycosis is a chronic and slowly progressive granulomatous disease caused by the filamentous Gram-positive anaerobic bacteria *Actinomyces*. These bacteria are frequently found in the oral cavity, gastrointestinal and female genital tracts (1). Several clinical forms have been described depending on the affected site. However, the precise etiology remains elusive. (2, 3, 4). The most prevalent form of actinomycosis is the cervicofacial variant, accounting approximately for 50% of the cases with a male predominance of 1.5-3.1 times (3, 5). Periapical actinomycosis (PA) is an uncommon chronic infection of endodontic origin. It is manifested by a persistent and recurrent draining fistula in the periapical region. Histopathology exams may confirm the diagnosis, while, bacteriological investigations allow the identification of the species involved. The surgical excision of the lesion is the treatment of choice. The aim of the present case is to describe a rare case of PA occurring in an adult patient.

## CASE PRESENTATION

The patient B.S.S, aged 66 years old, presented with a swelling of the chin associated with an acute pain related to the mandibular right canine (tooth 43). His medical history revealed a type 2 diabetes, treated, since 5 years, with Glucophage® 1 tablet/day. The blood glucose level was 1.5g/l and his blood pressure was 14/8. The patient was well monitored with a well-controlled diet, poor in sugar and free of salt. The exobuccal exam noted a located swelling of the chin. The endobuccal exam showed poor oral hygiene, and a painful swelling associated with a draining fistula related to tooth 43. Several prosthetic restorations (a bridge from 33 to 43, metal crowns), and mobile teeth were present (18, 15, 14, 25, 28). The panoramic radiography revealed a horizontal alveolar bone lysis and inadequate root canal treatments on multiple teeth (18, 15, 14, 25, 33, 42, 43). The retroalveolar radiography showed inadequate root canal fillings on teeth 42 and 43 associated with a radiolucent lesion (PAI>3) (figure 1). The marked radio opacity present in tooth 43 and at 2 mm from the apical portion of the canal indicated a fractured instrument. Both teeth were not mobile but tender to axial percussion. The diagnosis of a circumscribed cellulitis of the chin related to tooth 43 was established. Emergency treatment was conducted during the first session with root canal filling desobturation to allow the drainage of the pus through the access cavity on tooth 43 and the

draining fistula, as well. The fractured instrument near to the apical portion of the canal was bypassed. The wide and apparently centered canal was disinfected and obturated with Ca (OH)2 paste. Antibiotic mono therapy was prescribed (Amoxicillin® 2g/day for 7 days).

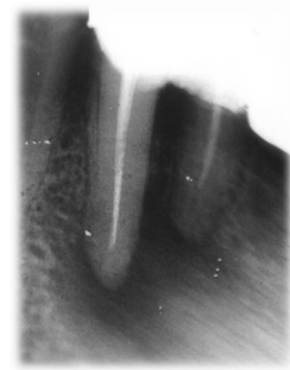


Figure 1

Preoperative radiography showing inadequate root canal. Filling in tooth 42 and 43, and a periapical radiolucent lesion

One week later, during the second session, the swelling was resolved and the draining fistula decreased in size. A tiny second canal was located, with a 10-K file at the buccal side of tooth 43. The retreatment of tooth 42 was performed in one session. All canals were mechanically prepared, disinfected with NaOCL 2.5% along with EDTA 17%, and filled using compacted warm gutta percha (System B, Obtura II), Thermafil was used to obturate the second buccal canal on tooth 43. As access cavities were performed through the metallic crowns, amalgam was used as a coronal restoration.

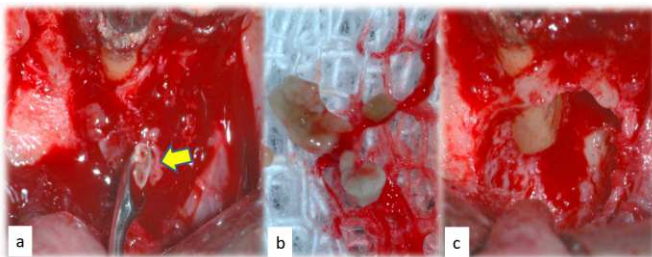
The outcome was marked by the persistence of the draining fistula and the extension of the periapical lesion (PAI>5) in tooth 43 (figure 2). Amoxicillin®, 2g/day was prescribed 2 days before the surgery. A surgical excision of the lesion was performed in the department of oral medicine and oral surgery. The periapical anesthesia with vasoconstrictor was performed, before the intra sulcular incision. The retraction of the surgical flap revealed a granulomatous infected tissue that perforated the buccal cortical bone. The excision of the large lesion showed pus associated with yellowish granules (figure 3 a and b). The resorbed apex of the canine was resected. The cavity was cleaned and the flap was sutured. The antibiotic treatment was prolonged for 5 days. The healing of the fistula and the periapical lesion marked the outcome (figure 4). Histological analysis (H and E staining) of the excised lesion and the purulent granules revealed a granulomatous tissue infected with actinomyces.

Special staining like, Grocott staining revealed the proliferation of the filamentous actinomycotic bacteria (figure 5 a and b). Bacteriological diagnosis at the species level was not performed.



**Figure 2**

Postoperative radiography after root canal retreatments in tooth 42 and 43, showing an increase in the size of the periapical lesion in tooth 43.



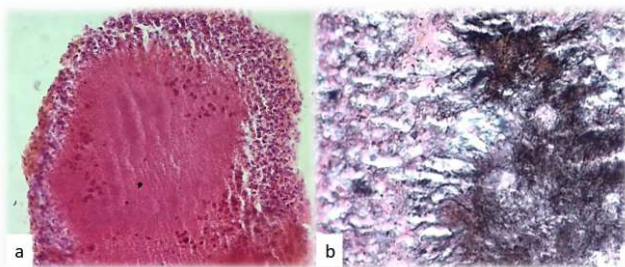
**Figure 3**

a: Surgical view showing cortical bone resorption, granulomatous lesion infected with granule pus (arrow); b: photo of the granule pus; c: clinical view of the cavity after excision of the granulomatous tissue.



**Figure 4**

Postoperative radiography showing healing of the periapical lesion after one year.



**Figure 5**

Histological view of the excised lesion ; a: actinomycetes granule surrounded by inflammatory immune cells (HE x 400), b: Black staining of actinomycetes granule with Grocott showing filamentous-shaped bacteria (Grocott x 400).

## DISCUSSION

Poor oral hygiene, periodontal diseases, dental caries and dental extractions may play a pivotal role in the development of PA (3, 5, 6). Indeed, some factors like immune impairment related-diabetes, head and neck radiotherapy, malnutrition and immunosuppression conditions may be considered as significant contributors for the PA occurrence (7). PA is the disease of young adults, with no gender predilection (8). Even not consistent, the most frequent affected site is the mandible (8, 9). As for symptoms, PA is usually asymptomatic but may cause swelling, painful abscess, fistula and purulent discharge. It may associate a necrotic pulp or an inadequate root canal filling (10). The radiographic appearance is not specific; it includes a radiolucent periapical lesion similar to what is described for granuloma and cyst of endodontic origin (11). In the present case, the lesion was a reactivated chronic abscess with purulent draining fistula, in a type 2 diabetic adult male patient who presented with a chin cellulitis. The lesion was persistent with an unfavorable outcome after adequate endodontic retreatment of tooth 43.

Taking into consideration the infrequency of the disease, histological analysis are underreported. In fact, less than 5% of the specimens undergo histological analysis (12, 13). Moreover, less than 1% of the estimated prevalence of PA was recorded (14). PA is characterized by the presence of sulfur granules with the Splendore-Hoeppli phenomenon, and inflammatory immune cells surrounding filamentous Gram-positive colony-forming aggregates. Tissue debris, antigen-antibody complex and fibrin are present. However, in some reports, actinomycotic colonies are lacking, despite the detection of *Actinomyces israelii*'s DNA in radicular cysts (14). There is no clear consensus regarding diagnostic criteria of PA. It is worth noting that *Actinomyces* has the ability to escape the immune system and grow as extraradicular biofilm even in the absence of an active root canal infection. Several techniques have been reported to identify the actinomycotic nature of the cervico-facial or the periapical infection, including anaerobic bacteria isolation and identification of the species, gas chromatography, immunohistochemistry and genetic and molecular techniques, as well (3, 14-19). However, some limitations are related to these techniques such as cost, complexity, accessibility and daily use in routine. In the case report, the presence of sulfur granules was obvious clinically,



suggesting a PA. histological analysis confirmed the presence of a granulomatous tissue surrounded by immune inflammatory cells, using H & E stain. The filamentous aspect of the bacteria was displayed using specific Grocott stain.

As for the treatment, it is recommended to perform an efficient disinfection of the necrotic canal or to undertake the retreatment of filled canals, in order to eliminate the intraradicular infection and make a final 3D obturation of the root canal system. The persistent extraradicular infection should be removed by a surgical excision of the periapical lesion. Dental extraction should be performed if indicated (2, 19). In symptomatic and acute conditions, antibiotic therapy may be considered. As often suggested, Penicillin is the first choice (3). As inadequate endodontic treatments were noted in the studied patient, retreatment was performed in both tooth 42 and 43. The persistence of the infection and the radiographic extension of the lesion lead to the indication of the surgical excision of the periapical lesion. Antibiotic treatment (Amoxicillin®) was administered to prevent reinfection and enhance the healing process in tooth 43. Favorable outcome was recorded after removal of the cause.

## CONCLUSION

Periapical actinomycosis is a chronic and a rare condition. The dentist should be informed about the ability of Actinomyces to form an extra radicular biofilm that escapes the immune system defenses. Elimination of the intra radicular infection by disinfection and adequate root canal filling should be associated with the surgical removal of the persistent periapical lesion.

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