Rare disease

Periapical actinomycosis

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Summary

Actinomycosis is a rare chronic infection caused by species of *Actinomyces* and characterised by abscess formation, tissue fibrosis, suppurative lesions and fistulas with purulent discharge containing sulphur granules. Owing to its multiform manners of presentation and non-specificity from its clinical features, it has been considered as a challenging diagnosis. Periapical actinomycosis is one of the rarest forms of actinomycosis occurring in the maxillofacial region. In its occurrence it presents in the form of persistent and recurrent draining fistula in the periapical region. We report a case of periapical actinomycosis occurred in endodontically treated teeth and accidentally found to be actinomycosis during histopathological examination. An insight towards the portal of entry of the organisms into the periapical region is also discussed. The practice of sending even a tiny bit of tissues to histopathology obtained from periapical surgery will very well demonstrate this disease and help in rapid resolution through appropriate antibiotic therapy.

BACKGROUND

Actinomycosis is a rare chronic infection caused by species of *Actinomyces* and characterised by abscess formation, tissue fibrosis, suppurative lesions and fistulas with purulent discharge containing sulphur granules. Owing to its multiform manners of presentation and non-specificity from its clinical features, it has been considered as a challenging diagnosis. Periapical actinomycosis is one of the rarest forms of actinomycosis occurring in the maxillofacial region. In its occurrence it presents in the form of persistent and recurrent draining fistula in the periapical region.

We report a case of periapical actinomycosis occurred in endodontically treated teeth and accidentally found to be actinomycosis during histopathological examination. An insight towards the portal of entry of the organisms into the periapical region is also discussed.

CASE PRESENTATION

A 30-year-old gentleman, with pain and pus discharge in the periapical region of lower central incisors persisting for 3 months, reported to our department. He underwent endodontic treatment 1 year before in both the lower central incisors in a different hospital. Intraoral examination revealed discoloured mandibular central incisors (figure 1) along with presence of sinus in the periapical region. Pus drainage was also noted through the sinus tract. Tenderness on percussion was present in both the central incisors.

An intraoral periapical radiograph was taken and it showed a 1×2 cm diffuse irregular radiolucency in the periapical region of both mandibular central and lateral incisors (figure 2). Teeth vitality test was carried out that showed both the lateral incisors to be vital. A provisional diagnosis of infected periapical granuloma was made.

As the radiograph reveals satisfactory obturation in both the central incisors periapical surgery was planned. Since the patient had draining sinus, it was decided to advise him empiric antibiotic therapy of amoxicillin 500 mg, 8 h for 5 days. The surgery included periapical curettage and apicoectomy in both the central incisors. During surgery, excessive destruction of bone especially in between the buccal and the lingual cortex noted and a mass of dirty white necrotic material was found and curetted that is quite unusual in routine periapical surgery. Curetted soft tissues were sent for histopathology examination.

INVESTIGATIONS

Postoperative examination of the specimen revealed multiple greyish white to greyish brown soft tissue bits amounting to 1 gm. Microscopically, sections showed granulation tissue with foci of necrotic acellular material with bacterial colonies. The background showed

Figure 1 Discoloured mandibular central incisors.

Figure 2 Radiograph showing diffuse irregular periapical radiolucency.

fragments of squamous epithelium and bone. The necrotic material is bordered by pinkish eosinophilic material (figure 3). The Splendore-Hoepli phenomenon (asteroid bodies) is found, which is the in vivo formation of intensely eosinophilic material (radiate, star-like, asteroid or club-shaped configurations) around microorganisms (fungi, bacteria and parasites) or biologically inert substances (figure 4).

On the basis of these presentations, actinomycosis was suspected and periodic acid Schiff (PAS) stain was done which clearly demonstrated multiple actinomycotic colonies (figure 5). Gram Brown-Brenn stain was also done and showed clusters of radiated, filamentous, branching, Gram-positive actinomyces microorganisms (figure 6). Gomori methenamine-silver nitrate stain demonstrated sulphur granules in the microbial colonies (figure 7).

Figure 3 H&E stain: foci of necrotic acellular material with bacterial colonies (×40).

Figure 4 Splendore-Hoepli phenomenon (×40).

Acid-fast staining was negative which can be used to distinguish *Actinomyces* spp., which are acid-fast negative, from *Nocardia* spp., which are variably acid-fast positive (figure 8).

DIFFERENTIAL DIAGNOSIS

Periapical abscess, periapical granuloma and periapical cyst.

OUTCOME AND FOLLOW-UP

A follow-up radiograph taken after 3 months showed healing bone defect (figure 9).

DISCUSSION

Actinomyces spp. are normal commensals of the human oropharynx, gastrointestinal tract and genitourinary tract. Six of 14 species have been implicated to be pathogenic in humans.^{1–3} *Actinomyces israeli* is most commonly incriminated in human disease.⁴ Since actinomycosis is a chronic, suppurative and granulomatous infection caused by anaerobic Gram-positive bacilli, it requires implantation into deep tissues. Puncture wounds, compound fractures and dental extractions are some of the possible routes of

Figure 5 Periodic acid Schiff stain: multiple actinomycotic colonies (×40).

Figure 6 Gram Brown-Brenn stain: clusters of radiated, filamentous, branching, Gram-positive actinomyces microorganisms ($\times 40$).

infection. The incidence of actinomycosis in cervicofacial is 55%, followed by 20% in abdominopelvic, 15% in thoracic and 10% in mixed organs, including skin, brain, pericardium and extremities.^{1-3 5}

Cervicofacial actinomycosis is further classified into central and peripheral types, of which the central variety is very rare in nature with the incidence of 1–2%.⁶ Periapical actinomycosis falls under central type which itself is very uncommon among central varieties.⁷

There is dearth of literature supporting the mode of entry of actinomycosis into the periapical region. However, it is believed that during root canal treatment the organisms are displaced from the oral cavity to the periapical region. Since majority of actinomycosis cases reported in the literature till date occurred in edodontically treated teeth which is in accordance with the above-mentioned hypothesis.⁷⁻¹⁰ Entry of these organisms through the periodontium can also be suspected, but in our case the gingival and the periodontal regions were apparently normal both clinically and radiologically.

Figure 7 Gomori methenamine-silver nitrate stain: sulphur granules ($\times 40$).

Figure 8 Acid-fast stain: negative ($\times 40$).

Another route of entry is endogenous in nature, which cannot be ruled out in all the cases. The belief that infection may be from endogenous sources is further confirmed by the fact that *A israeli* has never been demonstrated in soil, plants or any other object outside the body.¹¹

In our case amoxicillin was administered (500 mg orally every 8 h) for 2 weeks and the symptoms subsided uneventfully. *Actinomyces israeli* is sensitive to antibiotics that are effective against Gram-positive organisms such as penicillin, sulphonamides, streptomycin, tetracyclines, erythromycin and rifampicin, but high doses of long-term penicillin is the treatment of choice.^{12 13} A follow-up radiograph taken after 3 months showed healing bone defect (figure 9).

Figure 9 Follow-up radiograph: healing bone defect.

Learning points

- ▶ Owing to its multiform manners of presentation and non-specificity from its clinical features, periapical actinomycosis has been considered a challenging diagnosis.
- ▶ Periapical actinomycosis although rare should be included in the list of periapical pathology.
- ▶ Successful diagnosis and management of this case reaffirm the importance of examining every bit of tissue recovered from a lesion.

Competing interest None.

Patient consent Obtained.

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