REVIEW ARTICLE

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Periapical periodontitis complicated by cutaneous fistula of the cheek

Zapalenie tkanek okołowierzchołkowych zębów powikłane przetoką skóry policzka

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ABSTRACT: Inflammation of the periapical tissue may lead to the development of complications involving cooperation between many medical specialists. Cutaneous fistula is rare complication of chronic alveolus inflammation being diagnostic and therapeutic challenge due to unspecific symptoms. Correct diagnostic protocol may influence correct diagnosis, localization of the primary site of inflammation and the appropriate treatment.

KEYWORDS: Cutaneous Fistula, Dental Fistula, Abces

STRESZCZENIE: Stan zapalny w obrębie tkanek okołowierzchołkowych zębów może prowadzić do rozwoju powikłań wymagających leczenia przez zespół lekarzy różnych specjalności. Przetoka skórna jest rzadkim powikłaniem przewlekłego stanu zapalnego w obrębie zębodołu i niejednokrotnie stanowi wyzwanie diagnostyczno-terapeutyczne ze względu na brak specyficznych objawów. Odpowiednie przeprowadzanie procesu diagnostycznego może pozwolić na postawienie właściwej diagnozy, lokalizację pierwotnego ogniska stanu zapalnego oraz wdrożenie leczenia przyczynowego.

SŁOWA KLUCZOWE: przetoka skórna, przetoka zębowa, ropień

INTRODUCTION

Periapical periodontitis is usually caused by pathologic bacteria residing in the root canal of the tooth or as a result of previous endodontic treatment. The clinical course can be acute with severe pain, facial swelling and fever, or it may be chronic with non-specific symptoms. In most cases, complications are local, limited to the oral cavity. In some patients, the infection can spread outside the oral cavity leading to cutaneous fistulas, which can be challenging both diagnostically and therapeutically.

CASE REPORT

A 22-year-old female patient presented to the otolaryngology outpatient clinic due to an increasing inflammation of the left cheek. She complained about skin ulceration with purulent discharge for 6 months, located on the left cheek about 1.5 cm above the angle of the mandible. On examination, an eroded ulcer was noted about 1 cm in diameter covered with granulomatous tissue. The surrounding tissues were inflamed, hard, slightly erythematous, swollen and tender to palpation, without any obvious fluctuation. The inflammation, manifested by worsening edema and vasodilation around the wound with seropurulent discharge, tended to aggravate soon after cessation of oral antibiotics prescribed by successive physicians. After the initial oral empirical therapy with clindamycin, the effect was unsatisfactory and the patient was admitted to the laryngology department of another hospital. During her stay, the patient underwent head CT scan, which revealed in-

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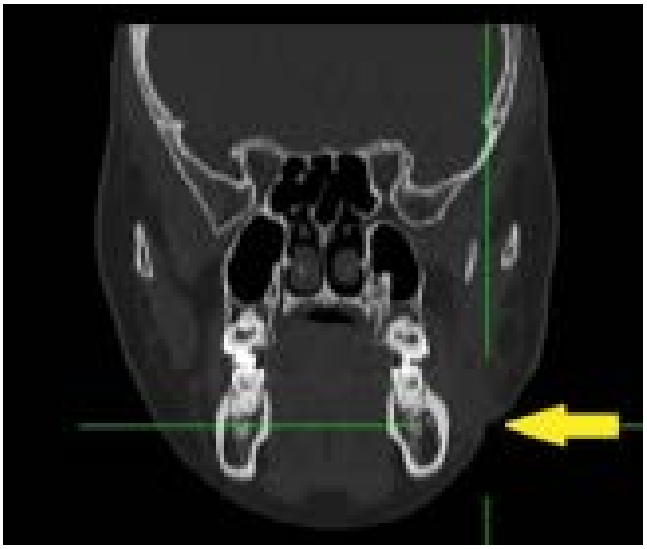

Fig. 1. Computed tomography scan in coronal section shows crater-like skin retraction of the left cheek.

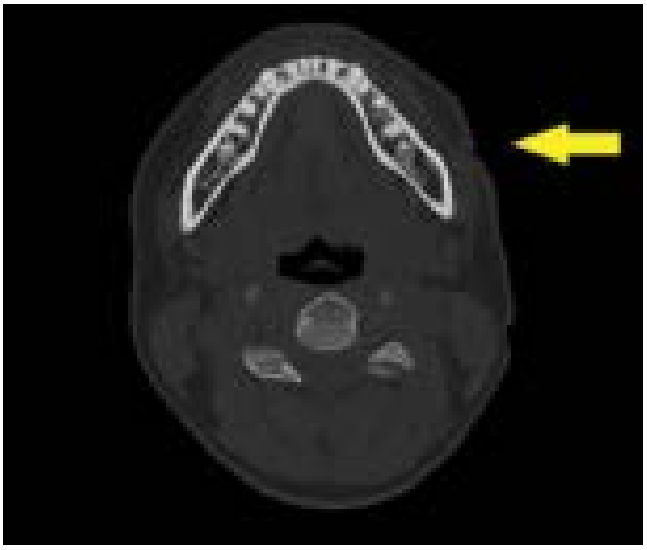

Fig 2. Computed tomography scan in axial section shows crater-like skin retraction and cortical layer obliteration of the anterolateral mandibular surface.

flammatory lesion within the soft tissues of the cheek. Cultures we collected and the wound was surgically managed. The cultures showed the growth of Gram-negative rods (*Prevotella* and *Escherichia coli*) and their sensitivity was determined. The targeted antibiotic was introduced and the wound decreased in size, however, without complete healing. The patient was treated for the next 6 months in ambulatory care setting by the dermatologist, laryngologist and infectious disease specialist. Because of progressing symptoms soon after termination of antibiotics, she received many courses of targeted therapy with oral penicillins, cephalosporins and lincosamides based on successive wound cultures. The patient denied any previous otolaryngologic treatment or chronic diseases. She reported that a year before she finished her orthodontic treatment, which had lasted several years.

On ENT examination, no abnormalities were found; normal dentition and oral mucosa advocated against acute inflammation. In ambulatory setting, ulcer cultures were collected revealing natural oral microflora (*Prevotella, Streptococcus oralis*), and the patient was qualified for admission for further surgical management. Also, tissue sample was collected and sent for pathology study, which later confirmed the inflammatory etiology. During hospitalization, laboratory tests and imaging were obtained, which showed no inflammatory marker elevation (white blood count 10.17 k/mm3; CRP 1.6 mg/L; procalcitonin < 0.2 ng/mL). On ultrasound, buccal abscess was excluded. The report only stated the presence of a hypoechogenic area, 7 x 8 x 10 mm in size, without blood flow on Color Doppler near the left angle of the mandible. The CECT was repeated showing cortical layer obliteration of the anterolateral mandibular surface over a short distance near the left angle of the mandible (Fig. 1 and 2). Because of a non-specific CT image, the decision was made to obtain an MRI scan, which showed abnormal subcutaneous soft tissue within the left cheek causing skin retraction, spreading over to the alveolar process near the left third molar; the presence of an abscess was excluded (Fig. 3 and 4). Due to suspected chronic periapical periodontitis of the mandibular left third molar (38) complicated by cutaneous fistula, the patient was consulted by the maxillofacial surgeon and qualified for tooth 38 extraction under local anesthesia followed by removal of the left buccal lesion under operating microscope to locate and resect the fistula canal. Postoperatively, the patient was given parenteral antibiotics based on the antibiogram, as well as antifungal treatment, leading to normal wound healing with satisfactory cosmetic outcome. The pathology study revealed skin covered with partially fibrotic granulomatous tissue with focal spots of mucous membrane, without any saprophytic colonies. During later observation in the outpatient clinic, the postoperative wound healed normally and no relapse of purulent discharge or ulceration was noted.

DISCUSSION

Cutaneous fistula is an extremely rare sequela of periapical periodontitis, posing diagnostic and therapeutic challenges because of its non-specific clinical course [1]. Local complications of periodontitis within the oral cavity are far more common, including periodontal, subperiosteal and submucosal abscess [2]. As illustrated by our case, cutaneous fistula is caused by chronic inflammation that is hard to recognize, leading to ca-

nal formation along the fascia and underneath normal oral mucosa. For a fistula to form, the process must involve tissues above the masseter muscle attachment to the zygomatic arch (in maxillary alveolar arch involvement) or below the masseter attachment to the mandible (in mandibular alveolar arch involvement) [3, 4].

Cutaneous fistulas resulting from chronic inflammation of the tooth and periodontium can be observed in patients without concomitant diseases or any acute symptoms, who are unaware of the ongoing processes; it can cause difficulties in making the right diagnosis and implementing appropriate surgical and medical treatment. It is estimated that even 50% of patients with odontogenic cutaneous fistulas undergo repeated surgical interventions, surgical biopsies and antibiotics without achieving local control of inflammation [5]. Periapical periodontitis can be caused by trauma, caries, tooth impaction, chronic pulpitis or periodontal diseases. However, inflammation can also be iatrogenic following previous dental interventions or oral surgery [6].

Non-healing or recurring facial skin lesions can be odontogenic, and therefore dental causes should be considered in order to eliminate the primary site of infection within the alveolar processes and teeth. Depending on lesion extent, presence of general symptoms and possible complications, conservative endodontic treatment may be applied or surgical dental extraction can be performed in the face of local or systemic sequelae [7]. The differential diagnosis should be based on excluding other local inflammatory conditions, which may not respond to standard antibiotics.

Firstly, single boil or carbuncle, active actinomycosis, granulomatous tissue or, in the case of orbital location, dacryocystitis should be excluded. Congenital malformations such as persistent thyroglossal duct or branchial cleft cyst can lead to permanent cutaneous fistula formation with episodic enlargement and purulent discharge. It is also important to exclude neoplasms by pathology study of tissue samples [2]. It should be mentioned that chronic skin lesions presenting as ulcers with intermittent seropurulent discharge can be a result of a salivary fistula in patients who had been subject to surgical treatment for salivary tumor or acute infection and did not comply with recommendations or were not properly followed up postoperatively [8]. The diagnosis should be based on basic laboratory test results and microbiology study in order to discern the etiology and clinical course of inflammation. The next key factor for right diagnosis is imaging workup to exclude soft tissue abscess and to assess maxillary and mandibular bone involvement (panoramic radiograph, ultrasound, CT scan). In selected cases, resonance imaging should be considered to evaluate soft tissues. The diagnostic workup should not delay dentist or maxillofacial surgeon consultation, which is crucial for final diagnosis and causal treatment.

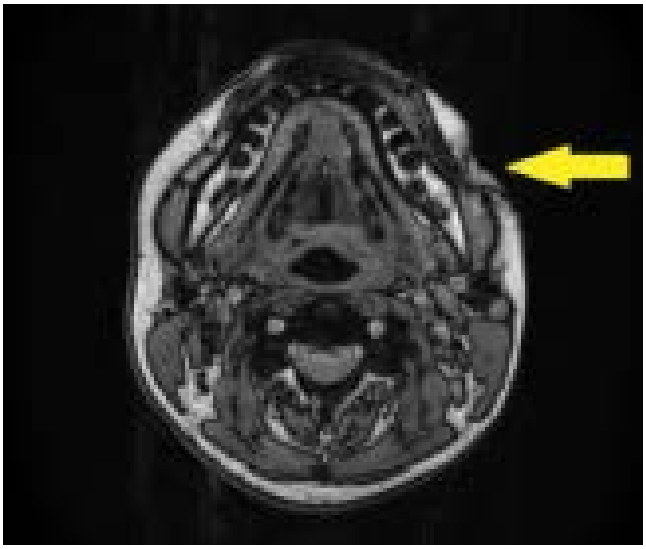

Fig. 3. Magnetic resonance scan in axial section shows crater-like ulceration covered with granulomatous and connective tissue.

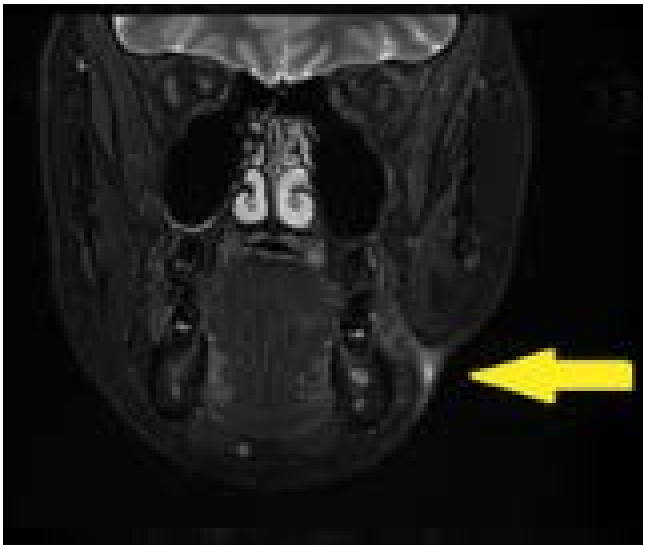

Fig. 4. Magnetic resonance scan in coronal section shows skin retraction with an area of contrast enhancement spreading from the lesion to the alveolar process of the mandible on the left.

CONCLUSIONS

Cutaneous fistula is a rare complication of periapical periodontitis, which poses diagnostic and therapeutic challenges due to its oligosymptomatic presentation. The key to right diagnosis and primary site infection elimination is the cooperation between medical specialists including the otolaryngologist, dentist and maxillofacial surgeon, accompanied by appropriate diagnostic plan. Dental and surgical treatment, together with antibiotics, can bring rapid improvement with a good cosmetic end result.

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