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Case Report

Limits of Conventional Treatment of Odontogenic Cutaneous Sinus Tract: A Case Report

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Abstract

Odontogenic Cutaneous Sinus Tract (OCST) is a rare and intriguing clinical entity that represents a pathway through the alveolar bone initiating at the apex of the infected tooth and vacates pus through the face or neck skin. A variety of etiologies can be involved in this pathology and its diagnosis is commonly challenging for dental and medical professionals as it can mimic other disorders. In this paper we report a case of odontogenic cutaneous lesion, evolving for a year, related to left mandibular canine which has a complex internal morphology. The limitations and potential pitfalls of clinical examination and imaging techniques are discussed, highlighting the need for other approaches to achieve accurate and definitive diagnoses. Furthermore, understanding the diverse microbial flora involved in OCST is crucial to devise targeted and effective therapeutic strategies especially when to indicate endo-surgical treatment and also antibiotic prescription.

Introduction

Odontogenic Cutaneous Sinus Tract (OCST) is a rare entity [1-3]. It represents a pathologic channel that initiates in the oral cavity and opens externally at the cutaneous surface of the face or neck [4-7]. Generally, this pathology is associated with longstanding infectious processes [2,4], considered as a common manifestation of pulpal necrosis with periapical pathosis [1], also, trauma, dental implant complications, salivary gland lesions, and neoplasms are causes of oral cutaneous fistulas [6].

Despite the fact that this condition is well documented, it still remains commonly misdiagnosed as it can mimic other disorders such as granulomatous disorder, basal cell and squamous cell carcinoma, salivary gland and duct fistula, infected cyst, furuncle, or actinomycosis like reported in our case [1,8,3,7].

For successful treatment of odontogenic cutaneous sinus tracts, the main approach should be to cut off communication between the infected area and the skin [10]. Usually, this can be done by non-surgical root canal treatment, but some cases require surgical-endodontic therapy in order to heal [1,9,7].

The aim of this article is to report a case of odontogenic cutaneous lesion related to left mandibular canine treated with combined surgical and endodontic treatment. It highlights the importance of a well-conducted therapy for the healing of this entity.

Case presentation

A healthy 62-year-old male patient visited the department of dentistry at “Sahloul University Hospital” in Sousse (Tunisia) for a cutaneous sinus tract that bothers him aesthetically. The patient reported that one year ago he consulted a dermatologist

who prescribed him antibiotics therapy for 2 months without regression of the lesion. Then, he was referred to a dentist office where he received an endodontic treatment on left mandibular canine, but the lesion didn't disappear.

Extraoral examination revealed a cutaneous lesion on submental region measuring 2 cm in diameter with depression aspect, indurated adherent plaque of discharging pus, mucoid material, and blood (Figure 1a). Clinical intra-oral examination revealed a poor oral hygiene, moderate fluorosis (Figure 1b). Tooth 33 was sealed coronary with a temporary filling (Cavit), asymptomatic, non-tender on percussion, and no deep pockets were present. Transillumination revealed a vertical coronary crack that extends from the cusp to the collar of the tooth (Figure 1c). The cord-like tissue was not palpable. Intraoral periapical radiograph revealed a well-circumscribed periapical radiolucency in relation with tooth 33 and insufficiency in root canal filling (Figure 2a). Mesial angulation radiograph revealed a missing second canal that was filled with sealer in its entrance (about 2 mm) (Figure 2b). The tracing of the sinus tract with gutta-percha cone wasn't possible because the orifice of the lesion was closed.

The patient presented an old panoramic radiograph dating one year before the root canal treatment, and it clearly reveals the internal morphology of tooth 33 with 2 canals and presence of the same periapical radiolucency (Figure 2c).

Following these examinations, diagnosis of pulpal necrosis with chronic peri-radicular periodontitis and extraoral cutaneous sinus tract related to 33 was made. Therefore, endodontic re-treatment was planned.

During the first visit, following the application of a rubber dam, removal of temporary coronal filling was done, and access opening was rectified with endo access bur. The gutta-percha removal was achieved with retreatment rotary files (ProTaper Retreatment Files) without the use of solvents. Then, to free access to the lingual canal diamond-coated ultrasonic tips were used for the removal of this hard material under dental operating microscope (Figure 3a). Finally, the two canals were visible and accessible (Figure 3b) and preparation with rotary files (Fanta Dental Rotary Files) was initiated with abundant irrigation 5.2% sodium hypochlorite. The working length was then determined (Figure 4) and calcium hydroxide-based medication mixed with Sodium chloride 0.9% solution was applied. In this radiography we can see a bone resorption located 3 mm below the corono-radicular junction.

During the second visit, complete canals preparation was done, and $Ca(oh)_2$ medication mixed with Sodium chloride 0.9% solution was reapplied due to serous fluid in the canal. At the third visit, the cutaneous lesion had different aspect becoming bigger, productive, budding with yellowish filaments emerging from its surface, sign of superinfection (Figure 5a,5b). At this point, the differential diagnosis of Cervicofacial actinomycosis was evocated and its confirmation required needle aspiration of the fistula to recover Actinomyces species from an appropriately cultured specimen. The lesion was disinfected and by seeing closer satellite cysts were visible (Figure 6). The puncture of the lesion did not extract enough pus or usable blood for microbiological examination so histopathological examination was necessary (Figure 7a) and biopsy of the lesion was scheduled. Multiple sections of a biopsy specimen from different tissue levels of the sinus tract were examinated and showed to not contain Actinomycosis colonies (Figure 7b). Prescription of antibiotics (Penicillin) was needed, root canal filling was postponed and re-cleaning and shaping of canals and appliance of $Ca(oh)_2$ were required.

At the fourth visit, after three weeks, signs of superinfection disappeared and the cutaneous sinus tract regresses with no discharge from its surface (Figure 8), thus, root canal filling was done using single-cone method and bioceramic sealer (Bio-Root™ RCS) (Figure 9).

After one-month, the tooth was clinically asymptomatic, but no signs of healing were noted, and the fistula didn't regress. So, endodontic surgery with fistulectomy was decided to assure the curettage of the peri-radicular lesion and the excision of the cutaneous sinus tract.

To access to the lesion, full-thickness flap was reflected, it revealed a fenestration on the vestibular bone situated in the peri-apical lesion covered by a granulous lesion. Two bone resorptions recovered by granulation tissue were noted on the midline between tooth 33 and tooth 32; one was located 3 mm below the corono-radicular junction like as observed in retro-alveolar radiograph and the other was more apical (Figure 10a). Curettage of granulation tissue was made followed by localized hemostasis, root-end resection at 3mm level, preparation with ultrasonic tip (Figure 10b,10c,10d) and finally root-end filling with Mineral Trioxide Aggregate (Figure 11) .

The patient was recalled after 1 week for final restorations. The coronal track was sealed with flow composite. After 3 months of the surgery, obvious signs of healing of the cutaneous sinus tract were observed and the prior depression aspect decreased leaving a scar measuring 4 mm in diameter (Figure 12). The patient may have to undergo a scar revision for esthetic reasons.

Figure 1: Clinical examination: **(a)** Extraoral examination revealed cutaneous lesion on submental region measuring 2 cm in diameter with depression aspect, indurated adherent plaque of discharging pus, mucoid material, and blood, **(b)** intra-oral examination revealed a poor oral hygiene, moderate fluorosis, **(c)** Transillumination revealed a vertical coronary crack that extends from the cusp to the collar of tooth 33.

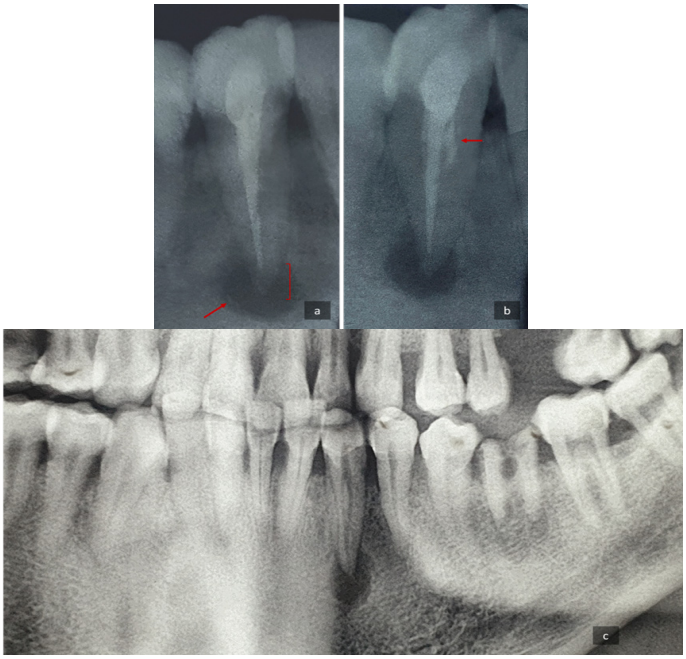

Figure 2: Radiographic examination: **(a)** Intraoral periapical radiograph revealed a well-circumscribed periapical radiolucency in relation with tooth 33 and insufficiency in root canal filling, **(b)** Mesial angulation radiograph revealed a missing second canal that was filled with sealer in its entrance (about 2 mm), **(c)** panoramic radiograph reveals the internal morphology of tooth 33 with 2 canals and presence of periapical radiolucency.

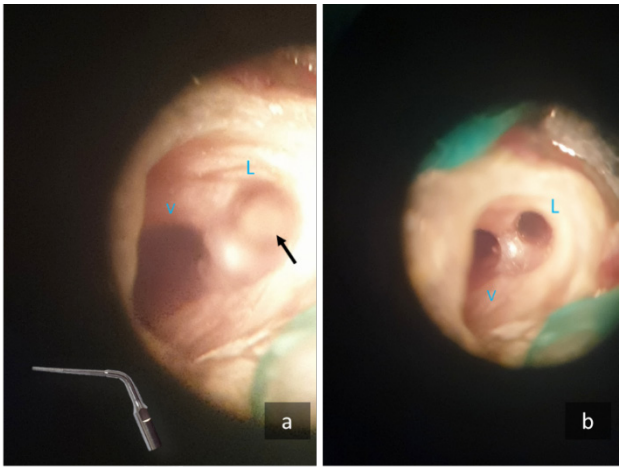

Figure 3: Access cavity view under dental operating microscope: **(a)** hard material (sealer) blocking access to lingual canal was removed with ultrasonic tips, **(b)** access cavity with two canals visible and accessible.

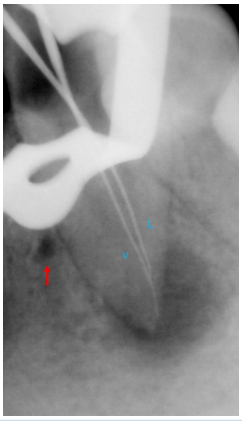

Figure 4: Per-operative radiograph: Determination of working length with files n°15.

Figure 5: Clinical extra-oral examination at third visit: **(a)** the cutaneous lesion became bigger, productive, budding with yellowish filaments emerging from its surface **(b)**.

Figure 6: Lesion aspect after disinfection: Presence of satellite cysts producing pus.

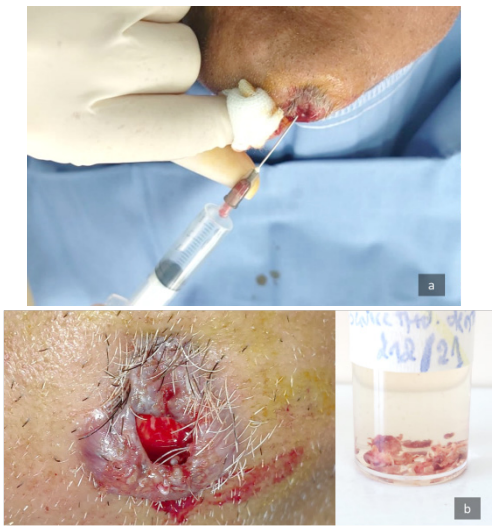

Figure 7: Complementary biological examinations: **(a)** Puncture of the lesion: Did not extract enough pus for microbiological examination, **(b)** histopathological examination.

Figure 8: Lesion aspect at fourth visit: signs of superinfection disappeared and the cutaneous sinus tract regresses with no discharge from its surface.

Figure 9: Post-operative radiograph: Root canal filling using single-cone method and bioceramic sealer (BioRoot™ RCS).

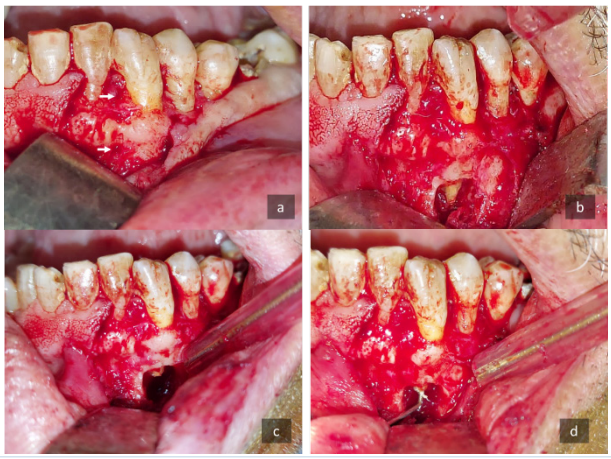

Figure 10: Surgical treatment: **(a)** Full-thickness flap was reflected and two bone resorptions recovered by granulation tissue were noted on the midline between tooth 33 and tooth 32; one was located 3 mm below the corono-radicular junction, **(b)** Curettage of granulation tissue and localized hemostasis, **(c)** root-end resection at 3mm level, **(d)** root-end preparation with ultrasonic tip and root-end filling with Mineral Trioxide Aggregate.

Figure 11: Post-operative radiograph: Root-end filling with Mineral Trioxide Aggregate (MTA).

Figure 12: Extraoral examination after 3 months: Signs of healing of the cutaneous sinus tract and the prior depression aspect decreased leaving a scar measuring 4 mm in diameter.

Discussion

An odontogenic cutaneous sinus tract is a pathway through the alveolar bone which initiates at the apex of the infected tooth and vacates pus through the face or neck skin [3,4]. It is commonly considered as consequence of suppurative process of a periapical abscess [1,2]. The sinus tract follows a path of least resistance and travels through bone and soft tissue [2,7,11]. Once the cortical plate has been perforated, the sinus tract's exit point is determined by local factors such as host resistance and anatomic arrangement of neighboring musculature and fasciae, the position of the tooth in the dental arch, the thickness of the bone and also factors such as gravity and the virulence of the microorganisms involved can play a role [7,4,5]. In fact, odontogenic cutaneous sinus tracts, rather than intraoral sinus tracts, are likely to occur if the apices of the teeth are superior to the maxillary muscle attachments or inferior to the mandibular ones. It was demonstrated that the prevalence of OCST varies from isolated case reports to 14.7% in large reported series [5] and mandibular teeth are most frequently associated with this pathology [11] like described in this case report.

The successful treatment of cutaneous sinus tract of dental origin depends on the diagnosis of the source which may be very challenging because; the patient may not have any apparent dental symptoms; only half of all patients ever recall having had a toothache, the lesion does not always arise in close proximity to the underlying dental infection and it often have a clinical appearance similar to other facial lesions, such as osteomyelitis, basal cell and squamous cell carcinoma, furuncles, bacterial infections, congenital fistulas, and pyogenic granulomas [8,4,7].

Clinically the orifice of OCSTs might extend from 1-20 mm in diameter and may present different shapes, it commonly resembles a furuncle, a cyst, an ulcer, a nodulocystic lesion with suppuration or it looks like a retracted or sunken skin lesion [7,12]. If misdiagnosed patients may undergo many inappropriate surgeries and courses of antibiotics before a definitive diagnosis is made and an appropriate therapy are initiated [4].

For the origin of oral cutaneous lesion, the traditional diagnostic approach is an invasive method based on tracing X-ray after the insertion of a lacrimal probe or sharp-tipped wire into the orifice opening until resistance is felt. This procedure damages the tissue's lesion and causes discomfort of the patient and stress of the operator [11], that's why authors prefer confirming the odontogenic origin of the lesion by tracing the sinus tract to its origin with gutta percha cones [8,7]; in our case we couldn't use this technique because the orifice of the cutaneous sinus

tract was closed. Other diagnosis tools are of critical importance such as: Negative pulp vitality testing which indicates the necrotic causal tooth and palpation of the involved area which often reveals a cord like track around suspected tooth [8,10], nevertheless, in most cases the epithelium lining the sinus tract does not extend deeper from the surface opening and may not be palpable [2] like in our case. In addition, periapical and panoramic films are essential for diagnosis by showing periapical radiolucency around the suspected tooth [8]. Some authors showed that CBCT imaging is an effective assistant diagnostic tool to confirm odontogenic etiology of cutaneous sinus tract; it reveals periapical radiolucency areas that are not visible upon panoramic and periapical radiography and cortical plate perforation leading to the lesion [11].

When it's adequately treated, closure of OCST may occurs within 5 to 14 days or few weeks [7,11]. Al-Kandari reported completely healing of the sinus tract after proper root-canal treatment without surgical treatment in three months leaving a small scar [8]. In this context, a non-healing lesion could be attributed to a non-odontogenic origin or inappropriate endodontic treatment [11]. In our report, we faced the second situation where tooth 33 was mistreated a year ago with insufficiency in filling of the principal canal and missing out the treatment of the second one.

Above all, it is crucial to know and understand the internal morphology of root canals for successful non-surgical as well as surgical endodontic therapy [13]. Over the literature, root canal morphology and configuration of mandibular canines have been well documented. Usually, these teeth have single root and single canal 87% but in 10% of cases, have two canals join at the root apex and in 3% have completely separated two canals [14,15]. There are several methods for investigating the root canal morphology: Cross-sectioning, microscopy, conventional radiography, Cone-Beam Computed Tomography (CBCT), micro-Computed Tomography (micro-CT) and clearing and staining methods [14,13]. CBCT and micro-CT are the two most recently introduced investigation methods [13] and researchers have showed that CBCT is a reliable tool in assessment of root canal and apical topography in mandibular canines, however it does not provide images that are as high resolution as those of micro-CT and its use in accessory canal detection is not recommended [13,14]. In our case, panoramic and retro-alveolar radiography used in different angulations were sufficient to visualize the internal morphology of 33 before starting the treatment. Based on the classification systems by Briseño-Marroquín and al, Vertucci, and Weine and al [13], the configuration of our dental case corresponds to Briseño-Marroquín's 2-2-1/1 also known as Vertucci's II or Weine's II (2-1). In general, some studies have attributed these variations to the role of genetics, the importance of ethnic background in tooth morphology and the difference in age and gender of patients [14].

Due to this unusual morphology, endodontic re-treatment was challenging especially when detecting the missing canal and removing the bioceramic sealer in its entrance which could not be achieved without the use of dental operating microscope and ultrasonic tip. Once canals were accessible cleaning and shaping were initiated. From a histological point of view, researchers demonstrated that teeth with chronic apical abscesses and sinus tracts have an overly complex infectious pattern in the apical root canal system and periapical lesion with a predominance of biofilms [2], for this reason, root-canal irrigation is a critical step on the success of the treatment. Authors showed that conventional chemical debridement with antimicrobial irrigants, such as sodium hypochlorite (NaoCl) or chlorhexidine do not always suffice to predictably render root canals free of bacteria [16]. Recently, endodontics lasers have been introduced as adjunctive antimicrobial procedures to raise the success of endodontic treatment and retreatments [1]. In fact, several studies claim that many biofilms are susceptible to Photodynamic Therapy (PDT) and 810 nm diode laser [1,16] and their use in canal disinfection reduced the CFU/ml. These two techniques did not show statistically significant differences and because of lower side effects, PDT could be the preferred technique [16].

Additionally, the use of calcium hydroxide as an intracanal medication is advocated for its benefits; eliminates bacteria that remain after mechanical debridement due to its high alkalinity and stimulates bone repair and participates in rapid and successful treatment of sinus tract associated with necrotic teeth [3,11]. In our case, $Ca(Oh)_2$ was renewed three times due to the presence of serous fluid in the canals. Every time it was mixed with saline which, according to authors, limits the dissolution of calcium hydroxide. Using polyethylene glycol (PEG) as a solvent, rather than water or saline, can increase the release of hydroxyl ions enhancing antimicrobial actions, and other improvements in performance and biocompatibility [17-20].

Between these sessions, the changes in the aspect of the cutaneous sinus tract becoming productive, budding with yellowish filaments emerging from its surface prompted us to make the differential diagnosis of Actinomycosis. Indeed, this lesion is characterized by a granulomatous inflammation which form multiple abscesses connected by sinus tracts that may discharge with a typical thin, watery characteristic "sulfur granules" [18,19,21]. These granules are an important diagnostic marker of Actinomyces species as it contains masses of filamentous organism. Macroscopically, it resembles yellow grains of sand (0.1-1 mm in diameter) but can become dark brown due to the deposition of calcium-phosphate [21]. The diagnosis of this lesion is usually confirmed by culturing the organism, for at least 14 days by needle aspiration of an abcess[18]. In our case, this procedure wasn't feasible so, curettage of the fistula and histological examination was necessary, and the result showed to be negative. At this point, antibiotics (Amoxicillin and Metronidazole) were prescribed to the patient. Following the American Association of Endodontists, in cases of odontogenic cutaneous sinus tracts, antibiotics are indicated to prevent secondary infections and bacteremia in systemically unhealthy cases with fever, malaise, lymphadenopathy, progressive diffuse swelling, and trismus [10,17,9]. Otherwise, systematic antibiotic therapy will result in a temporary reduction of the drainage and apparent healing. However, this tract will recur immediately after the antibiotic therapy is completed unless the initial source is not eliminated [8,4].

The obturation of root canal system was performed using single-cone method with Bioceramic endodontic sealer (BioRoot™ RCS), it exhibits unique physiochemical properties that can provide exceptional outcomes [3]. In 2021, a novel root canal filling technique, known as ultrasonic Vibration and thermos-hydrodynamic obturation (VibraTHO) was introduced. It incorporates indirect ultrasonic sealer activation and short-range warm vertical compaction of a single Gutta percha cone. This technique is almost as fast and user-friendly as the conventional single-cone technique and can be a more effective root canal filling method for anatomically complex root canal systems [22,23].

Studies that reported a high success rate for non-surgical treatment of teeth with sinus tracts proved that closure of the tract and periradicular tissue healing may mostly rely on how effectively the clinician controls the intraradicular infection [2]. In our case, periradicular surgery was decided to remove the granulation tissue and to improve on the result of the treatment. Indeed, cutaneous sinus tracts are usually lined with granulomatous tissue with a lumen containing a purulent exudate [8].

This pathology usually heals by forming a small pit and hyperpigmentation, which decrease over [9]. Nevertheless, in certain cases surgical removal of the sinus tract extra orally may be necessary [10] especially when a residual scar persists [11,16] and it proved to be an adjunct for prompt and speedy management of the lesion [3].

For the Prognosis of this odontogenic sinus tract, it has a good one after proper treating the offending tooth and surgical management of the extraoral lesion. Even so, more follow-up visits are required to confirm the success of our treatment.

Conclusion

This report highlights the importance of correct diagnosis and therapy in cases of odontogenic cutaneous sinus tracts. By enhancing the understanding of this challenging condition, it is hoped that early recognition and appropriate treatment approaches can be adopted, leading to improved patient outcomes and enhanced quality of life.

The success of the management of this pathology depends on proper root canal treatment of the causal tooth which can be followed by surgical therapy. Beforehand, knowledge of the internal root canal morphology of the tooth is crucial for the success of the treatment. Indeed, through this case we showed that despite its rarity, the presence of extra canals in mandibular canine should be explored before starting the root canal treatment and its missing will lead to failures.

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