CASE REPORT

Odontogenic cutaneous sinus tract - misdiagnosis and follow-up in a seven-year-old boy

Qianyi Deng[1,2], Mustapha Rammal[3], Huini Zhang[1,4], Hongwen He[4], Fang Huang[1,4*]

[1] Department of Pediatric Dentistry, Guanghua School of Stomatology, Hospital of Stomatology, Sun Yat-sen University, Guangzhou, China
[2] Paediatric Dentistry and Orthodontics, Faculty of Dentistry, the University of Hong Kong, Hong Kong SAR, China
[3] St-Lukes-Roosevelt Hospital, Department of Dentistry, New York, USA
[4] Guangdong Provincial Key Laboratory of Stomatology, Guangzhou, China

Abstract

Cutaneous sinus tracts of dental origin are relatively rare, but frequently misdiagnosed. In this case report, we present a seven-year-old patient with a cutaneous lesion in the left submandibular region misdiagnosed by a physician as an abscess secondary to suppurative lymphadenitis, and thus incorrectly treated with surgery and systemic antibiotics. Following a detailed dental examination, the patient was correctly diagnosed with an odontogenic sinus tract from a periapical abscess of tooth 36. Treatment of the immature tooth was initiated with apexification combined with nonsurgical endodontic treatment. The cutaneous and the periapical lesions were all resolved after the treatment and there has been no recurrence during an eight-year follow-up.

Keywords: cutaneous sinus tract, cutaneous fistula, odontogenic, apexification, immature permanent tooth

*Corresponding Author
Email address: hfang@mail.sysu.edu.cn
Tel: +86 20 83862553
Fax: +86 20 87330582

Introduction

Odontogenic cutaneous sinus tracts (OCSTs), uncommon and therefore frequently misdiagnosed, are generally caused by chronic endodontic infections (Chen *et al.*, 2016; Chouk & Litaiem, 2020). Patients presenting with OCSTs are often treated incorrectly with systemic antibiotics or surgical procedures by physicians because the dentition is often asymptomatic. When the infection persists, patients are often referred to a dental clinician to determine a dental etiology (Gimenez-Garcia *et al.*, 2015; Yadav *et al.*, 2014). Necrotic teeth are frequently overlooked because the offending tooth manifests little symptoms after perforation of the periosteum. In addition, the stoma of the sinus tract may be located at a distant site from the infection, for instance, at a submental cutaneous sinus tract of mandibular second molar origin, making the diagnosis very challenging (Bai *et al.*, 2014; Ghazali & Ngeow, 1996; Ong & Ngeow, 1999). Moreover, the variable symptoms of OCSTs also lead to diagnostic confusion and mismanagement (Lee *et al.*, 2016).

The mechanism of cutaneous sinus tract formation depends greatly on anatomic pathways. Odontogenic infection spreads from the apex of the necrotic tooth and penetrates the alveolar bone and soft tissues through the path of least resistance, limited only by muscle attachments and fascial planes (Kaban, 1980). As a result, OCSTs may occur when the infection is superior to the maxillary attachment of the buccinator muscle and inferior to the mandibular attachments of the buccinator, mylohyoid, and mentalis muscles (Laskin, 1964). Odontogenic infections in children and adolescents are theoretically easier to develop into extraoral sinus tracts because teeth are often partially erupted with immature roots and the alveolar process is less developed than in adults, with a higher chance of the infection to cause a perforation beyond the muscular attachments (Swales *et al.*, 2016). However, OCSTs in children are infrequently reported.

This article presents a case of a cutaneous sinus tract misdiagnosis that was successfully treated as an endodontic periapical infection with nonsurgical root canal therapy of the left mandibular first molar (tooth 36) in a seven-year-old male patient, with an eight-year follow-up.

Case report

On clinical examination, a cutaneous lesion in the left submandibular region was noted. The patient's medical history was examined, which indicated that two months prior to the referral, the patient was diagnosed with suppurative lymphadenitis subsequent to swelling with pain in the left submandibular region and odynophagia with fever. His physicians commenced treatment with incision and drainage along with a systemic antibiotic regiment of erythromycin ethyl succinate 100mg/2.5ml, four times daily. The symptoms disappeared and the incision healed after two weeks. However, a month later, swelling reoccurred with intermittent discharge of pus. Incision and drainage was repeated along the lesion, which presented as an abscess of size 11× 8mm. Nevertheless, the lesion was not resolved in a month, and discharge of pus still persisted (Figure 1A, 1B).

A seven-year-old Chinese boy was referred to our clinic at the Department of Pediatric Dentistry, Guanghua School of Stomatology, Hospital of Stomatology, Sun Yat-sen University, Guangzhou, P.R. China. His chief complaint was of “severe pain lasting two days on the lower left side when chewing”.

An intraoral clinical examination revealed a slight discoloration in the mesial marginal ridge of the left mandibular first molar (tooth 36). The tooth was positive to percussion and pain on biting, however tested negative to vitality and cold tests. A panoramic radiograph was taken, which revealed a radiolucent lesion on the mesial aspect of the crown of tooth 36 that extended to the mesial pulp horn, while the occlusal aspect of the crown was intact (Figure 1C). An intraoral periapical radiograph showed an extended periapical radiolucency at the distal root of tooth 36, along with immature apices (Nolla stage 9) (Figure 1D). From the radiographic findings, a diagnosis of periapical abscess with secondary cutaneous sinus tract formation was made.

Treatment was commenced with standard access cavity preparation by removal of caries under rubber dam on tooth 36. The four canals: mesiobuccal (MB), mesiolingual (ML), distobuccal (DB), and distolingual (DL) canals were cleaned and shaped. There was discharge of pus, and the tooth was left open for drainage. Two days later, on the follow-up visit, there were no signs of acute inflammation. A rubber dam was placed after administration of an inferior alveolar nerve block using 2% Lidocaine 1:100,000

epinephrine and further canal debridement was performed. A non-setting calcium hydroxide paste (LELE®, Shanghai Eryi & Zhangjiang Biomaterial Co., Ltd., Shanghai, China) was applied as an intracanal antimicrobial dressing for two weeks. On the third visit, the patient was asymptomatic and healing of the cutaneous sinus tract had occurred (Figure 2A). Apexification treatment was initiated by applying a premixed calcium hydroxide paste with iodoform (Vitapex®, morita, Osaka, Honshu, Japan) to the root canal system (Figure 2B). Between appointments, the coronal cavity was provisionally sealed with glass ionomer cement (Fuji IX GP®, GC Co., Tokyo, Japan).

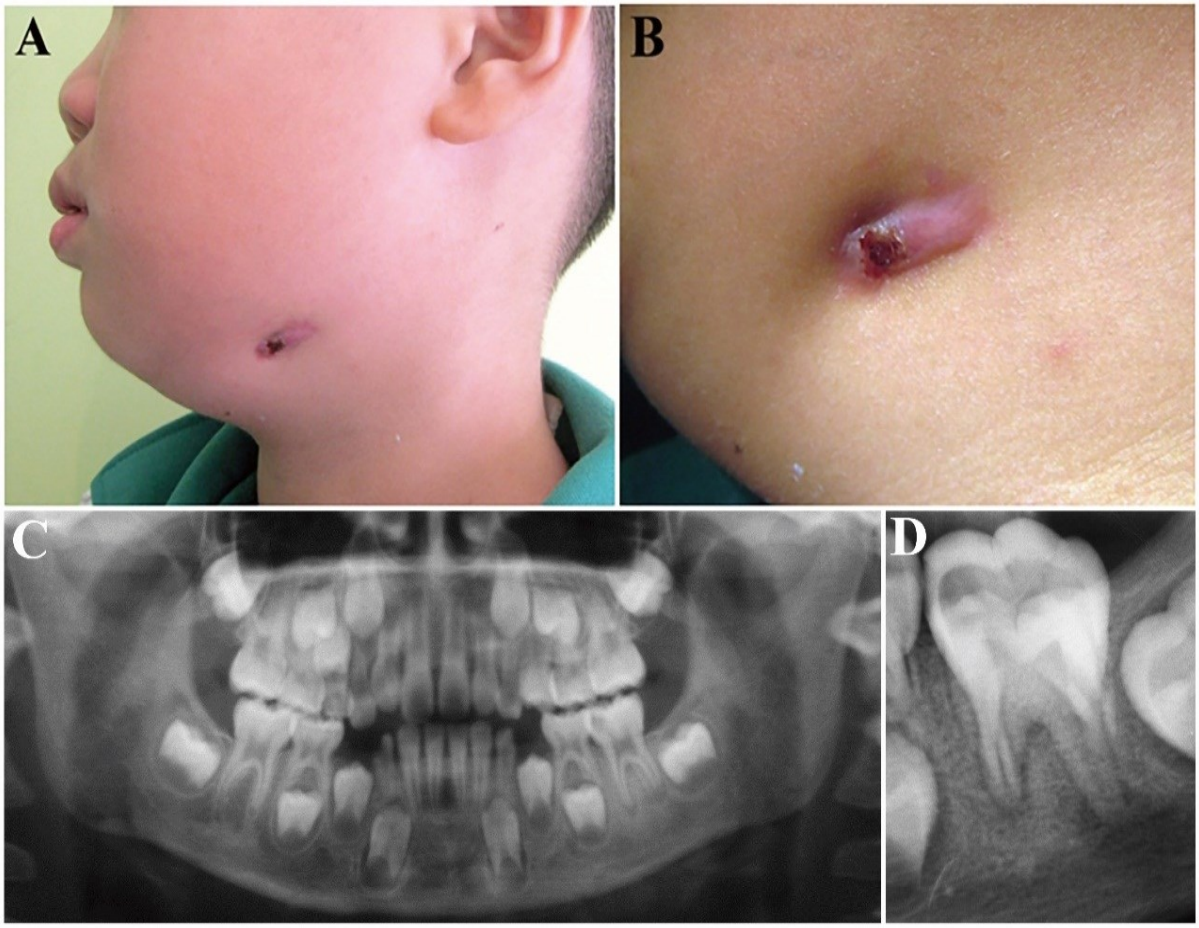

Figure 1. (A) Pre-operative view of the cutaneous sinus tract. Repetitive drainage and antibiotic therapy did not resolve the condition. (B) Magnified view of the cutaneous sinus tract. (C) Pre-operative panoramic view, showing a radiolucent lesion on the mesial aspect of the crown of tooth 36 extending to the mesial pulp horn, while the occlusal aspect of the crown was intact. (D) Periapical radiograph of tooth 36, revealing immature apices and an extended periapical radiolucency at the distal root.

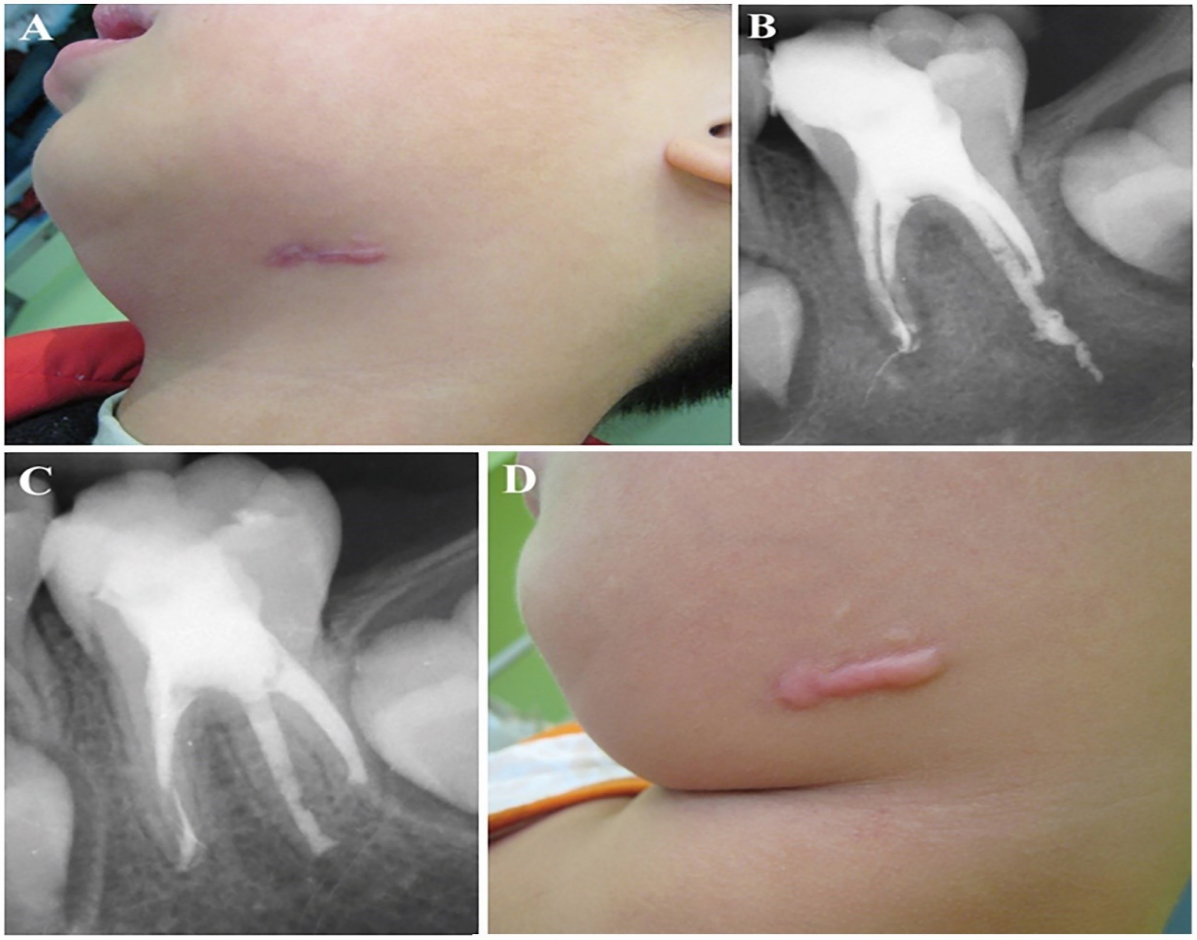

Figure 2. (A) After 2-week intracanal dressing with calcium hydroxide paste, healing of the cutaneous lesion had occurred. (B) Radiograph after applying a premixed calcium hydroxide paste with iodoform to the root canals. (C) Radiograph showing resolution of the periradicular radiolucency, formation of the calcified barrier at the mesial root apices and initiation of apexification. (D) Extraoral photograph after 7 months, showing hypertrophic scar formation.

Radiographic examination after seven months demonstrated resolution of the periradicular radiolucency and the emergence of a calcified barrier at the mesial root apices (Figure 2C). An extraoral view at this time exhibited the formation of a hypertrophic scar (Figure 2D). Eleven months after the apexification treatment, a periapical radiograph demonstrated the absorption of the root canal dressing (Figure 3A). Nickel-titanium rotary files (ProFile®, Dentsply Maillefer, York, PA, US) were used through the application of a crown down technique to shape the MB, ML, DB, DL canals to a size #35.04. Concurrently, all the canals were irrigated copiously with 2.5% sodium hypochlorite, 3% hydrogen peroxide, and normal saline, respectively. The canals were obturated using Cortisomol sealer (Morita, Osaka, Honshu, Japan) by way of a warm gutta-percha technique. A final postoperative radiograph revealed a homogeneous and dense root filling (Figure 3B). The access opening was restored with composite resin.

After an eight-year follow-up, the tooth was visibly unremarkable, with no radiographic and clinical symptoms (Figure 3C). The cutaneous lesion resolved with minimal scarring, except for a slight hyperpigmentation of the area (Figure 4A, 4B).

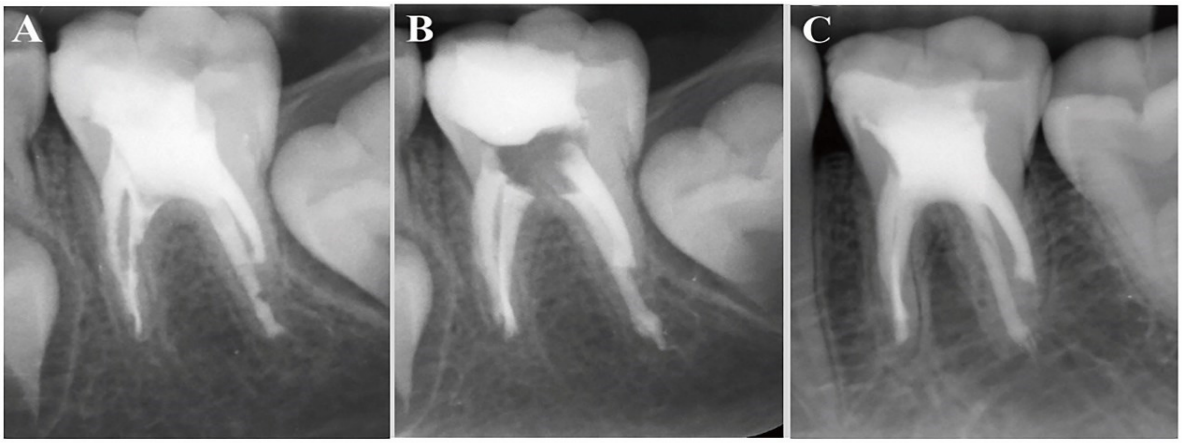

Figure 3. (A) Radiograph 11 months after apexification treatment, presenting the absorption of the root canal dressing. (B) Post-operative radiograph of tooth 36, revealing homogeneous and dense root filling. (C) Radiographic follow-up after 8 years.

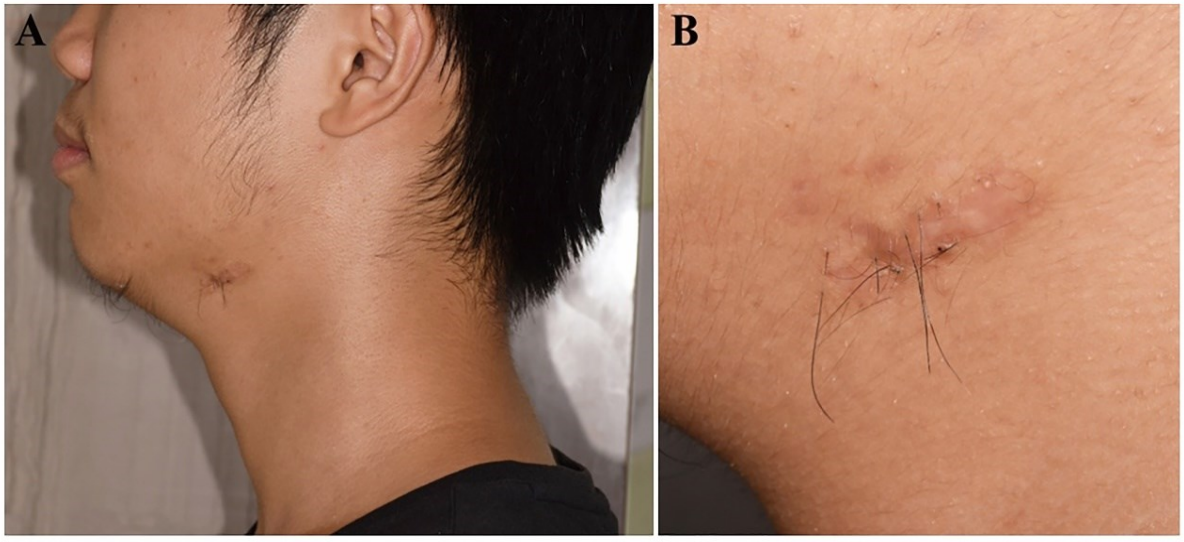

Figure 4. (A) Clinical examination follow-up after 8 years. (B) Magnified view of the area.

Discussion

In this case, based on the patient's age and radiographic examination, the enamel calcification of tooth 36 was incomplete. As permanent teeth erupt, an additional period of two years is needed to complete the calcification process by exposure to saliva, thus the dentition is susceptible to caries formation during this time (Chin *et al.*, 2016). In addition, acute caries occurs frequently at this age, which progress faster and can consequentially cause endodontic infection. Acute carious lesions often present as light brown or grey in color, penetrating from the occlusal or interproximal aspect leading to early diagnostic challenges. Histologically, the region of the sinus tract constitutes a conglomerate of inflammatory cells and granulation tissue. The luminal region is comprised of granulomatous tissue along with purulent exudates containing polymorphonuclear leukocytes and chronic inflammatory cells. After treatment is initiated with root canal therapy or extraction, healing begins, resulting in minimal scar formation (Yi *et al.*, 2017).

As reported in the literature, OCSTs are frequently misdiagnosed and as a

consequence often mismanaged (Bashar *et al.*, 2019; Swales *et al.*, 2016). In addition, differential diagnosis is paramount in making an efficient constructive treatment plan. In this case report, differential diagnosis should include traumatic lesion, epidermal cyst, furuncle, carbuncle, pyogenic granuloma, salivary gland fistula, and actinomycosis (Cantatore *et al.*, 2002; Gulec *et al.*, 2001; Jamshidi *et al.*, 2015; Tavee *et al.*, 2003). Therefore, detailed history taking and comprehensive examination is crucial in determining an accurate diagnosis. Furthermore, dental and medical clinicians should be aware that often odontogenic lesions can be a common cause of cutaneous sinus tract infection. Once the etiology is clear and the causative factors are removed, the stoma and the sinus tract will close spontaneously within several days.

In this report, tooth 36, which was determined to be nonvital and immature, was successfully treated with apexification using calcium hydroxide to induce formation of a calcified barrier at the root terminus. The time needed to form a barrier of a root canal apex varies, which is dependent upon the size of the apical foramen, as well as the severity of the periapical lesion.

Alternatively, mineral trioxide aggregate (MTA) can be used as a rapid apexification protocol. However, both MTA and calcium hydroxide do not promote continued development of the root, leading to a thin root dentin and a large canal lumen, thus predisposing the tooth to possible fractures (Cvek, 1992). Recent developments in revascularization and regeneration therapy in apexogenesis to yield thicker and longer root development using 3Mix-MP paste (including ciprofloxacin, metronidazole, minocycline with propylene glycol, and macrogol) have shown to be an effective protocol for management of immature permanent molar teeth with pulpal necrosis (Sonmez *et al.*, 2013).

Furthermore, when accessing healing, the duration of lesion plays a critical role. Bodner *et al.* evaluated cases of 28 children with odontogenic cutaneous sinus tracts and found rapid healing occurs in lesions with shorter duration, whereas extraoral scarring occurred in lesions with longer duration (Bodner *et al.*, 2012). In the case report, the duration of the origination of the lesion up to the date of the correct diagnosis was estimated to be less than 3 months and the lesion healed with minimal scarring. Moreover, location of the lesion should also be taken into consideration. As presented in our case, the lesion was located in the submandibular region with relaxed skin tension and the scar is parallel to existing skin-tension lines, consequently yielding a better condition for wound remodeling (Son & Harijan, 2014).

Conclusions

Patients exhibiting non-healing cutaneous sinus tracts in the maxillofacial region need a thorough medical and dental evaluation. Once the infection of the offending tooth is removed by endodontic treatment or tooth extraction, healing of the cutaneous lesion will occur. Early diagnosis and accurate treatment are paramount for healing of OCSTs without scar formation.

Acknowledgements

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References

Bai, J., Ji, A. P., & Huang, M. W. (2014). Submental cutaneous sinus tract of mandibular second molar origin. *International Endodontic Journal,* 47(12), 1185–1191.

Bashar, A. K. M., Akter, K., Chaudhary, G. K., & Rahman, A. (2019). Primary molar with chronic periapical abscess showing atypical presentation of simultaneous extraoral and intraoral sinus tract with multiple stomata. *BMJ Case Reports,* 12(9), e229039.

Bodner, L., Manor, E., Joshua, B. Z., Barabas, J., & Szabo, G. (2012). Cutaneous sinus tract of dental origin in children - a report of 28 new cases. *Pediatric Dermatology,* 29(4), 421–425.

Cantatore, J. L., Klein, P. A., & Lieblich, L. M. (2002). Cutaneous dental sinus tract, a common misdiagnosis: a case report and review of the literature. *Cutis,* 70(5), 264–267.

Chen, K., Liang, Y., & Xiong, H. (2016). Diagnosis and treatment of odontogenic cutaneous sinus tracts in an 11-year-old boy: a case report. *Medicine,* 95(20), e3662.

Chin, J.R., Kowolik, J.E., & Stookey, G.K. (2016). Dental caries in the child and adolescent. In: Dean, J.A. (ed.), *McDonald and Avery's Dentistry for the Child and Adolescent: Tenth Edition.* St. Louis, MO: Elsevier, pp. 155–176.

Chouk, C., & Litaiem, N. (2020). Oral Cutaneous Fistula. In *StatPearls [Internet]*. Treasure Island (FL): StatPearls Publishing. Updated 16 May 2020, from http://www.ncbi.nlm.nih.gov/books/NBK539822/

Cvek, M. (1992). Prognosis of luxated non-vital maxillary incisors treated with calcium hydroxide and filled with gutta-percha. A retrospective clinical study. *Endodontics & Dental Traumatology,* 8(2), 45–55.

Ghazali, M.N., & Ngeow, W.C (1996). Unusual presentation of a sinus due to impacted lower third molar – A case report. *Dental Journal of Malaysia*, 17(1), 19–21.

Gimenez-Garcia, R., Martinez-Vera, F., & Fuentes-Vera, L. (2015). Cutaneous sinus tracts of odontogenic origin: two case reports. *Jounal of the American Board of Family Medicine,* 28(6), 838–840.

Gulec, A. T., Seckin, D., Bulut, S., & Sarfakoglu, E. (2001). Cutaneous sinus tract of dental origin. *International Journal of Dermatology,* 40(10), 650–652.

Jamshidi, D., Moazami, F., Sobhnamayan, F., & Taheri, A. (2015). Clinical and histopathologic investigation of periapical actinomycosis with cutaneous lesion: a case report. *Journal of Dentistry, Shiraz University of Medical Sciences,* 16(3 Suppl), 286–290.

Kaban, L. B. (1980). Draining skin lesions of dental origin: the path of spread of chronic odontogenic infection. *Plastic and Reconstructive Surgery,* 66(5), 711–717.

Laskin, D. M. (1964). Anatomic considerations in diagnosis and treatment of odontogenic infections. *The Journal of the American Dental Association,* 69, 308–316.

Lee, E. Y., Kang, J. Y., Kim, K. W., Choi, K. H., Yoon, T. Y., & Lee, J. Y. (2016). Clinical characteristics of odontogenic cutaneous fistulas. *Annals of Dermatology,* 28(4), 417–421.

Ong, S.T., & Ngeow, W.C. (1999). Median mental sinus in twins. *Dental Update*, 26(4), 163–165.

Son, D., & Harijan, A. (2014). Overview of surgical scar prevention and management. *Journal of Korean Medical Science,* 29(6), 751–757.

Sonmez, I. S., Oba, A. A., & Almaz, M. E. (2013). Revascularization/regeneration performed in immature molars: case reports. *Journal of Clinical Pediatric Dentistry,* 37(3), 231–234.

Swales, K. L., Rudralingam, M., & Gandhi, S. (2016). Extraoral cutaneous sinus tracts of dental origin in the paediatric patient. A report of three cases and a review of the literature. *International Journal of Paediatric Dentistry,* 26(5), 391–400.

Tavee, W., Blair, M., & Graham, B. (2003). An unusual presentation of a cutaneous odontogenic sinus. *Archives of Dermatology,* 139(12), 1659–1660.

Yadav, S., Malik, S., Mittal, H. C., & Puri, P. (2014). Odontogenic cutaneous draining sinus. *Journal of Craniofacial Surgery,* 25(1), e86–88.

Yi, J., Baek, I.H., & Min, K.W. (2017). Application of cone-beam CT for the diagnosis of an odontogenic cutaneous lesion near the nasolabial fold: a case report. *Hong Kong Journal of Dermatology & Venereology,* 25, 24–27.