

CASE REPORT

Submental cutaneous sinus tract of mandibular second molar origin**J. Bai¹, A.-P. Ji¹ & M.-W. Huang²**

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Abstract

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Aim To report a case in which a submental cutaneous sinus tract resulted from apical periodontitis associated with a mandibular second molar.

Summary A 53-year-old man presented with a chronic cutaneous sinus tract in the submental region, which had previously been misdiagnosed as a thyroglossal fistula. The origin of the sinus tract was shown by sinus tract angiography to be the left mandibular second molar (tooth 37), which had apical periodontitis. The sinus tract healed after extraction of the tooth and partial excision of the lesion from an extraoral approach. Histological examination confirmed that the tract was lined with granulation tissue and not with epithelial tissue. A submental sinus tract drainage pathway was observed. Prompt dental evaluation, especially of all mandibular teeth, should be considered when assessing cases of submental cutaneous sinus tracts.

Key learning points

- An odontogenic origin should be part of the differential diagnosis for orofacial skin lesions.
- Cutaneous sinus tracts of mandibular molar origin are complex and thus a comprehensive examination should be stressed.
- It is necessary to examine all mandibular teeth in cases of odontogenic submental cutaneous sinus tracts.
- Sinus tract angiography can be used to identify the sinus tract pathway and to confirm the associated teeth.
- The treatment of an odontogenic cutaneous sinus tract requires the elimination of the source of infection.

Keywords: cutaneous sinus tract, odontogenic, submental.

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Introduction

An odontogenic cutaneous sinus tract is a pathologic channel that originates in the oral cavity but exits at the cutaneous surface of the face or neck, commonly resembling a furuncle, cyst, ulcer or retracted, sunken skin lesion (Cantatore *et al.* 2002, Gupta *et al.* 2011, Samir *et al.* 2011). Because of its rare occurrence and the absence of associated symptoms in approximately half of the individuals affected, it is commonly misdiagnosed (Cantatore *et al.* 2002, Gupta *et al.* 2011), usually leading to inappropriate treatment (e.g. surgical excision, biopsy, radiotherapy and/or antibiotics) and eventual recurrence of the cutaneous sinus tract (Tidwell *et al.* 1997, Johnson *et al.* 1999, Mittal & Gupta 2004, Gupta *et al.* 2011, Abuabara *et al.* 2012). The chronic nature of the lesion results in patients typically having multiple appointments with medical practitioners prior to a correct diagnosis (Cantatore *et al.* 2002, Samir *et al.* 2011).

Odontogenic cutaneous sinus tracts are known to develop as a result of pulp infection, chronic apical periodontitis, root fracture, chemical irritation and dental trauma (Chan *et al.* 1997, Nakamura *et al.* 1999, Witherow *et al.* 2003, Mittal & Gupta 2004, Sheehan *et al.* 2005, Pasternak-Júnior *et al.* 2009, Samir *et al.* 2011, Herd *et al.* 2012). Without effective treatment, infection can spread from the necrotic pulp into the root apical area around the tooth, resulting in apical periodontitis (Cantatore *et al.* 2002, Samir *et al.* 2011). Seeking the path of least resistance, the purulent by-products of infection can then travel through the bone and the soft tissue (Kaban 1980, Al-Kandari *et al.* 1993, Tidwell *et al.* 1997, Güleç *et al.* 2001, Cantatore *et al.* 2002, Sheehan *et al.* 2005, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011).

This paper reports the case of a patient with an odontogenic midline submental cutaneous sinus tract, which was initially misdiagnosed as a thyroglossal fistula. Radiologic evidence revealed a drainage tract that originated from the left mandibular second molar and opened on the midline of the submental region. This particular tract path has not been previously reported in the literature. This case report serves as a reminder to dental and medical practitioners that inflammatory facial lesions can originate from distant dental infections.

Report

A 53-year-old man was referred to the Peking University School and Hospital of Stomatology complaining of a purulent discharge from a midline submental cutaneous sinus tract. Of 4 months prior to the referral, the patient presented with submental swelling, which was then diagnosed as an infection secondary to a thyroglossal duct cyst (Fig. 1a). Based on the diagnosis, the patient was prescribed antibiotics, but no improvement was observed. Subsequently, continuous purulent drainage penetrated the submental skin (Fig. 1b). After referral, the initial dental examination focused on the mandibular anterior teeth, which appeared healthy. Orthopantomographic imaging revealed periapical radiolucencies associated with the left mandibular second molar (tooth 37) (Fig. 1c). The patient was then admitted with an initial diagnosis of thyroglossal fistula. However, sinus tract angiography clearly showed that the cutaneous sinus tract originated from the root of tooth 37 and that the lingual bone of the tooth was resorbed (Figs. 1d and e). The patient stated that he had experienced pain on biting on the left mandibular teeth 7 months earlier and that the pain dissipated without treatment within one week. Upon further inspection, tooth 37 was extremely mobile and nonresponsive to heat and electrical pulp tests. Considering the medical history and examination results, the patient was finally diagnosed as having an odontogenic cutaneous sinus tract secondary to a necrotic pulp and asymptomatic apical periodontitis of tooth 37.

Tooth 37 was subsequently extracted, revealing a vertical root fracture. Curettage of granulation tissue was conducted in the socket of tooth 37, and bone resorption at the lingual side of the tooth was confirmed. Finally, the sinus lining was partially removed from the extraoral

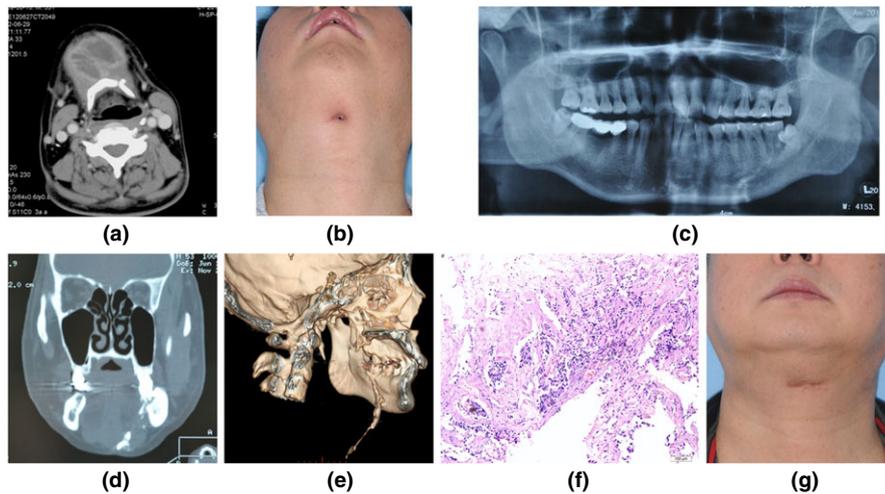


Figure 1 (a) Computed tomography (CT) image showing the initial lesion in the submental area, which appeared as an infection secondary to a cyst linked to the hyoid bone. (b) Extraoral view of the sinus tract in the submental area. (c) Orthopantomographic image showing a radiolucent area at the apex of the left second molar of the mandible with an impacted third molar. (d) CT image of fistula angiography, showing the contrast agent traced to a radiolucent area at the apex of the left second molar of the mandible and absorption of the lingual bone. (e) 3D CT image showing the contrast agent (sinus tract) traced to the apex of the left second molar of the mandible. (f) Pathological results showing granulation tissue and not epithelial tissue lining the tract. (g) Healed sinus tract area in the submental area.

aspect, and the opening was sutured to ensure a cosmetic result. Pathological analysis confirmed that the tract was lined with granulation tissue without epithelial tissue (Fig. 1f). Upon review, 3 months after the treatment, the cutaneous sinus tract had completely healed with minimal scarring (Fig. 1g).

Discussion

Due to the rarity of odontogenic cutaneous sinus tracts, misdiagnosis is common. Misdiagnosing this condition can lead to improper and unnecessary treatment that does little or no good for the patient, in addition to increasing the chronicity of the lesion.

Pathogenesis

Nearly 80% of reported cases of odontogenic cutaneous sinus tracts have been associated with mandibular teeth (Hodges *et al.* 1989, Foster *et al.* 1992, Tidwell *et al.* 1997, Esteban Palacio *et al.* 1999, Tavee *et al.* 2003, Witherow *et al.* 2003, Mittal & Gupta 2004, Sheehan *et al.* 2005, Yasui *et al.* 2005, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011, Herd *et al.* 2012), half of which were anterior teeth producing sinus tracts on the mentum base (Tidwell *et al.* 1997, Witherow *et al.* 2003, Mittal & Gupta 2004, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011, Herd *et al.* 2012). Fig. 2a shows the common sinus tract opening positions reported in the literature. Cutaneous sinus tracts originating from premolars tend to open in the mandibular area (the inferior border of the cheek) (Pasternak-Júnior *et al.* 2009, Samir *et al.* 2011). For diseased mandibular molars, the extraoral opening sites vary but they are most often located in the mandibular, submandibular or cervical areas and rarely found in the retromandibular area (Cioffi *et al.* 1986, Nakamura *et al.* 1999, Mittal & Gupta 2004, Yasui *et al.* 2005, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011, Samir *et al.* 2011, Abuabara *et al.* 2012). Of these cases, the pathway through the buccal cortical plate towards the mandibular area is the most common (Nakamura *et al.* 1999, Yasui *et al.* 2005, Pasternak-Júnior *et al.*

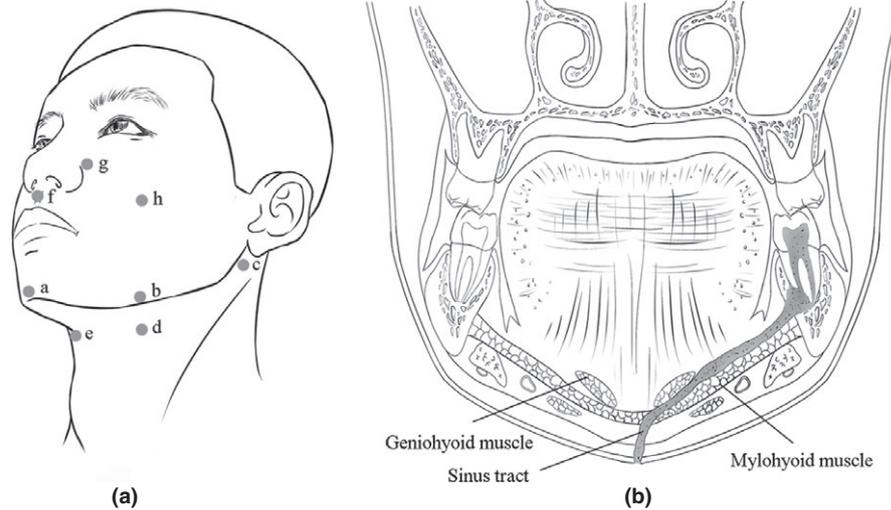


Figure 2 (a) Common sinus tract opening positions reported in the literature. (a: on the mentum base, b: mandibular area [inferior border of the cheek], c: retromandibular area, d: submandibular area, e: submental area, f: the philtrum or upper lip region, g: nose, nasolabial fold and infraorbital region, h: upper part of the cheek). (b) Possible drainage passage of the submental cutaneous sinus tract of left mandibular second molar origin.

2009, Gupta *et al.* 2011, Samir *et al.* 2011). When purulent drainage penetrates the lingual cortical plate, the extraoral sinus tract often opens in the submandibular or cervical areas (Cioffi *et al.* 1986, Mittal & Gupta 2004, Gupta *et al.* 2011, Abuabara *et al.* 2012). Cutaneous sinus tracts associated with the maxillary anterior teeth are likely to open on the philtrum, the upper lip region, nose, nasolabial fold or infraorbital region (Cohen & Eliezri 1990, Tidwell *et al.* 1997, Güleç *et al.* 2001, Witherow *et al.* 2003, Sheehan *et al.* 2005, Peermohamed *et al.* 2011), whereas those associated with the maxillary posterior teeth are likely to open on the upper part of the cheek (Mittal & Gupta 2004, Gupta *et al.* 2011).

This paper is the first report of a midline submental cutaneous sinus tract caused by a mandibular second molar. After analyzing computed tomography (CT) images of the patient and considering anatomic factors, a possible drainage tract was suggested (Fig. 2b). The observations, together with the literature on this topic, demonstrate the complexity of mandibular molar drainage pathways and the importance of the careful examination of mandibular molars during the investigation of lesions.

Diagnosis

Cutaneous sinus tracts may result from several diseases, such as furuncles, bacterial infections, carcinomas, osteomyelitis, pyogenic granulomas and congenital fistulas. However, an odontogenic cutaneous sinus tract should be considered to be an important part of the differential diagnosis (Tidwell *et al.* 1997, Johnson *et al.* 1999, Mittal & Gupta 2004, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011, Abuabara *et al.* 2012). It is important that dental and medical practitioners be aware that any facial and cervical cutaneous lesion could have a dental origin (Mittal & Gupta 2004, Sheehan *et al.* 2005, Gupta *et al.* 2011). However, the diagnosis of an odontogenic cutaneous sinus tract is also challenging because these lesions do not always arise in close proximity to the underlying dental infection and because many patients do not exhibit dental symptoms (Held *et al.* 1989, Sheehan *et al.* 2005, Gupta *et al.* 2011, Samir *et al.* 2011).

To make the correct diagnosis, obtaining the clinical and medical history, especially of any past episodes of dental pain, from the patient is important (Nakamura *et al.* 1999). Pulp tests

and periradicular diagnostic testing should be performed on any suspected teeth and adjacent teeth (Gupta *et al.* 2011). Due to the complexity of the drainage pathways, all homolateral teeth (mandibular or maxillary) of the cutaneous sinus tract should be examined, especially in cases where lesions lie in the midline area. Furthermore, the attending clinician must search carefully for a potential odontogenic infection, which could be chronic, having been present for months or years (Cioffi *et al.* 1986).

Radiographic findings are also important for the diagnosis and identification of affected teeth (Nakamura *et al.* 1999, Abuabara *et al.* 2012). Radiolucencies at the apex of a tooth, shown by panoramic or periapical radiography, could be a sign of infection at the site (Sheehan *et al.* 2005). Although conventional panoramic radiographs might be useful for identifying the location of suspected teeth, confirming which tooth is associated with a cutaneous draining sinus tract is difficult, especially when multiple teeth are suspected. Some studies have suggested inserting a probe or an endodontic gutta-percha point through the sinus tract to obtain radiographs so as to aid the identification of the affected teeth (Nakamura *et al.* 1999, Pasternak-Júnior *et al.* 2009, Gupta *et al.* 2011). However, because this method does not appear suitable for the assessment of cutaneous draining sinus tracts from distant teeth, it was not employed in this case. Thus, sinus tract angiography was used to identify its pathway and to confirm the diagnosis.

Finally, some studies have also suggested conducting microbiologic culturing and sensitivity tests of the sinus tract exudate to identify microbial flora and to exclude specific infections, such as syphilis and actinomycosis (Gupta *et al.* 2011).

Treatment

The treatment of odontogenic cutaneous sinus tracts requires the elimination of the source of infection, either by root canal treatment in the case of a restorable tooth or by extraction in the case of a nonrestorable tooth (Chan *et al.* 1997, Nakamura *et al.* 1999, Sheehan *et al.* 2005, Samir *et al.* 2011). These lesions are refractory to a single treatment with antibiotics (Witherow *et al.* 2003, Sheehan *et al.* 2005, Samir *et al.* 2011). Surgical excision of the lesion without appropriate treatment of the infected teeth will inevitably lead to recurrence (Witherow *et al.* 2003). The necessity of surgical excision is controversial. Some studies have recommended complete excision of the sinus tract lining (Gupta *et al.* 2011), whereas others have contended that surgical treatment and antibiotic therapy are not necessary following dental treatment (Cioffi *et al.* 1986, Johnson *et al.* 1999, Nakamura *et al.* 1999, Mittal & Gupta 2004, Sheehan *et al.* 2005). Furthermore, some controversy exists regarding whether these tracts are lined by epithelium. An increasing number of related studies in the literature have shown that such tracts are generally lined by granulation tissue without epithelium (Cioffi *et al.* 1986, Johnson *et al.* 1999, Nakamura *et al.* 1999, Sheehan *et al.* 2005).

In this case, tooth 37 was extracted, and the sinus tract opening was sutured after partial resection from the extraoral aspect. The fixed biopsy of the tract was embedded in paraffin, and complete serial histological sections were stained with haematoxylin and eosin. The result of histological examination confirmed the presence of granulation tissue and the absence of epithelial tissue. The patient was not prescribed antibiotics. 3 months after treatment, the sinus tract had healed without recurrence. This result confirmed the initial misdiagnosis and the existence of a rare and unusual drainage pathway.

Conclusion

This case highlights the significance of including odontogenic origins to the differential diagnosis of orofacial skin lesions. Although odontogenic cutaneous sinus tracts most frequently arise in close proximity to the cause of the underlying infection, the possibility of a distant

dental cause must also be considered, especially when examination of the adjacent teeth shows no obvious odontogenic pathology. In those cases of extraoral sinus tracts in the submental region, examining all the patient's mandibular teeth is important and necessary. A detailed dental examination must be performed for the mandibular molars due to the complexity of the drainage pathways associated with these teeth.

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