



Case report

An uncommon cause for a non-healing cutaneous fistula in the parotid gland area - A case report

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ABSTRACT

Introduction: Orocutaneous fistulae of dental origin are uncommon but well documented in the literature. This condition is often misdiagnosed because of the multiplicity of manifestations and the atypically presentation of the condition. Dental symptoms are rare. This makes diagnosis and treatment a challenging process.

Presentation of a case: A 67-year-old patient presented in the emergency room with an abscess in the parotid area. After incision and drainage the patient developed a non-healing fistula in the region of the parotid. Multiple treatment attempts and several investigations did not solve the problem. Finally, a panoramic x-ray showed a hidden, infected and displaced tooth in the right mandibular angle. After surgical extraction of the tooth and fistula excision, healing was uneventful and there was no recurrence.

Discussion: Given the diagnostic challenge, the real origin of the condition is frequently only discovered after several unnecessary interventions that may have harmed the patient. The key to early diagnosis is dental examination and dental radiographs. The goal of treatment is the elimination of the infection by surgical extraction of the tooth or non-surgical endodontic therapy.

Conclusion: Early diagnosis and rapid treatment minimize patient discomfort and reduce the probability of further complications. The differential diagnosis of sinus tract of dental origin should be considered in any case of unclarified non-healing skin lesion in the head and neck area, especially if initial treatments have failed. An early interdisciplinary approach is essential.

1. Introduction

Cutaneous sinus tracts of dental origin are an uncommon manifestation of dental infection that is often misdiagnosed [1]. Precise data on prevalence are not found in the literature. The similarity of the clinical presentation with other diseases such as basal cell carcinoma, squamous cell carcinoma, osteomyelitis, congenital fistula, salivary gland fistula, infected cyst, mycotic infection, pyogenic granuloma, actinomycosis and foreign body reaction [2] often leads to inadequate management. The reason is the distance between the source of the disease and the manifestation site and the frequent lack of toothache. Incorrect diagnosis often leads to multiple unsuccessful attempts at incision, biopsy and drainage. Even the exaggerated use of antibiotics [3], radiotherapy [4] and electrodesiccation [5] have been reported. Approximately 80% of cutaneous dental sinus tracts arise from mandibular teeth with almost half of these lesions involving anterior mandibular teeth [6]. The

most important source is pulpal necrosis with periapical abscess formation due to caries or dental trauma. Cutaneous sinus tracts of odontogenic origin appear more commonly in the submandibular or submental region as nodulocystic suppurative lesions [7,8]. The manifestation site is often not in the proximity of the infected tooth but arises after pathogens have slowly formed a track through the cancellous bone following the path of least resistance to perforate the cortical plate and present either intra-, or extraorally. To provide the correct diagnosis dental examination, dental radiography, vitality testing and probing the fistula are essential. The foremost aim of treatment is to eliminate the source of infection. Treatments of choice are root canal treatment or surgical extraction [9].

A particularly challenging situation presents when the tooth is completely covered by the oral mucosa. We report a case in which an orocutaneous fistula of dental origin was managed empirically as a recurrent parotitis because of the similarity of symptoms. This case has

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been reported in line with SCARE (Surgical case report) criteria [10].

2. Presentation of a case

A 67-year-old woman presented at the emergency unit with an increasingly painful swelling and reddening in the area of the right parotid gland that had persisted for one week. A previously initiated empirical antibiotic therapy with Augmentin® (amoxicillin/clavulanate potassium, GlaxoSmithKline AG, Münchenbuchsee, Switzerland) orally for 7 days had been unsuccessful. On admission there was general malaise with elevated temperature and clearly elevated blood inflammatory parameters. The clinical examination showed an incipient weakness of the mouth branch of the facial nerve on the right side. The patient also suffered from increasing trismus. The intraoral inspection showed an edentulous patient with a total upper and lower denture. An ultrasonographic examination revealed fluid collection with signs of abscess formation in the parotid area. The case history showed two similar episodes in the past. At that time, a recurrent sialadenitis of the parotid gland had been assumed. The treatment with a short antibiotic regime was successful. The patient was generally healthy without regular medication. An empirical intravenous antibiotic therapy with Augmentin® (amoxicillin/clavulanate potassium, GlaxoSmithKline AG, Münchenbuchsee, Switzerland) was started. The submandibular incision and drainage of the abscess in the area of the lower parotid gland lobe was performed in general anaesthesia. Swelling and pus formation returned and so a repeat procedure was performed three days later. The wound went on to heal uneventfully after this procedure. The patient was discharged eight days after admission in good general condition.

A few days after dismissal from the hospital an exuding fistula in the area of the incision developed. The patient suffered with this condition for the following four months. All empirical and resistance-based attempts to treat this condition with antibiotics failed. A surgical approach that involved trying to stitch up the fistula was unsuccessful. Magnetic resonance imaging (MAGNETOM® Aera, 1.5T, Siemens Healthcare AG, Zürich, Switzerland) with sialography was performed. The findings did not reveal a possible reason for the persistent fistula. An additional sialendoscopy did not show any abnormalities.

Four months after discharge a panoramic x-ray of the mandible was taken. This image showed an impacted and displaced tooth in the region of the right mandibular angle. Bone radiopacity around the roots indicated osteolysis due to infection (Fig. 1). The patient was re-admitted to the Department of Maxillofacial Surgery at the same hospital. The revised diagnosis was now cutaneous fistula of dental origin with chronic infection of the third molar of the right lower jaw (Fig. 2). Empirical oral antibiotic therapy (Clindamycin®, Pfizer PFE Switzerland GmbH, Zürich, Switzerland) was started and the tooth was surgically removed in regional anaesthesia (Fig. 3). The fistula was excised. The healing process was uneventful. Antibiotic therapy was given for 7



Fig. 1. Panoramic x-ray showing the impacted and displaced tooth in the right mandibular angle (red arrow). The blue arrow shows periapical osteolysis indicating infection. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)



Fig. 2. Cutaneous fistula of dental origin in the area of the right lower parotid lobe and mandibular angle. Macerated skin with turbid secretion.



Fig. 3. Intraoperative picture after incision of the oral mucosa and discreet removal of mandibular bone. Crown of the displaced lower right wisdom tooth (black arrow).



Fig. 4. Clinical situation three months after the operation. Complete healing. Discreet scarring without any sign of recurrence.

days. Three weeks later healing was complete. There was no residual swelling, no pain and no recurrence of the fistula. At the three-month follow-up the result was still satisfactory without any signs of recurrence (Fig. 4).

3. Discussion

Cutaneous fistula of dental origin is and remains a diagnostic challenge. A periapical dental abscess secondary to caries is the major factor behind a large proportion of such cases [11]. Other causes are trauma and periodontal disease. Patients often present without obvious dental discomfort, which results in unfocused or inappropriate therapy, consequently, extending the time of infection [12]. In fact, after

bacterially induced necrosis of the pulpa and the formation of a sinus tract, pain or swelling in the environment of the tooth are rare. The sinus will drain cutaneously if the inflammation drains out of the jaw above the maxillary muscle attachments or below the mandibular muscle attachments [9]. The most common cutaneous exit points are the submental or the submandibular area. Odontogenic cutaneous sinus tracts have a soft, erythematous, and slightly depressed appearance [13]. Palpation of the surrounding tissue can result in fluid discharge draining through a central opening [9]. The most important tools for providing the correct diagnosis are the dental examination, vitality test, dental radiographs and careful history-taking especially because previous case reports have suggested that presentation with an odontogenic cutaneous sinus tract is more likely to occur in patients with a history of previous dental disease [3]. The goal of the treatment is to eliminate the source of infection. A previously reported review of 137 cases found that 106 (77%) were treated by extraction and 27 (20%) were solved by surgical or conservative nonsurgical endodontic therapy [14]. Some earlier studies suggested that the sinus tract is lined with epithelium and propose surgical extraction of the tooth to achieve healing [15]. Subsequent studies have demonstrated that the sinus tract is often lined with granulation tissue, indicating that it can be treated by nonsurgical root canal therapy [16,17]. The isolated excision of the fistula without eliminating the source of the infection will result in a recurrence of the condition. Antibiotics should be administered during treatment of the affected tooth to ensure complete resolution of the infection [13]. In contrast, other studies have suggested that if the primary source of infection is properly eliminated the lesion will heal spontaneously without the need for antibiotics [3,16].

In our case the infection was due to a hidden third molar. Since it was completely covered by the mucosa and there was partial impaction in the mandibular bone, the tooth was not visible at clinical examination. High tech imaging like Magnetic Resonance Tomography also failed to provide the correct diagnosis, probably because of the atypical presentation of the symptoms and the unexpected source of the condition. In this case dental panoramic radiography was found to be the most appropriate method to achieve correct diagnosis. Panoramic x-ray showed an osteolytic process around the root apex. This finding suggested a periapical dental abscess. Bacterial contamination of the hidden tooth probably originated in the oral cavity due to recurrent small mucosal injuries possibly caused by the lower dental prosthesis. In our case preservation of the tooth was not possible so we opted for surgery. Despite recent studies suggesting that the sinus tract and cutaneous lesion will heal without treatment once the primary odontogenic cause is removed [18], we believe that the additional removal of the fistula was necessary to achieve complete healing particularly where long duration (more than six months) of the sinus tract can be assumed (sinus tract probably lined with epithelium). An additional problem is the proximity of the fistula to the parotid gland. This circumstance can favour the formation of a parotid salivary fistula.

4. Conclusion

We conclude in summary that dental sinus tracts are often misdiagnosed and therefore inappropriately treated. The differential diagnosis of sinus tract of dental origin should be considered for any unclarified non-healing skin lesion in the head and neck area, especially if initial treatment has failed. An early interdisciplinary approach and cooperation between specialties will minimize patient discomfort, avoid unnecessary medical treatment, and reduce the likelihood of complications. The possibility of a dental source should be evaluated for any cutaneous lesion of uncertain origin in the facial area.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

The treatment plan was approved in the joint discussion by the maxillofacial surgeons, the otorhinolaryngologists and the radiologists.

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None.

Author contribution

The lead surgeons were G. Ghazal and A. Rabufetti. A. Husner was the caring otorhinolaryngologist who referred the patient. In addition, A. Rabufetti is responsible for concept and definition of intellectual content, documentation and manuscript writing. C. Leiggenger, M. Augello and A. Husner carried out manuscript editing and reviewing. The responsible physician is C. Leiggenger.

Conflicts of interest

None.

Registration of research studies

No registration because it's not a research study but a case report of an interesting disease.

Guarantor

A. Rabufetti / C. Leiggenger.

Consent

The patient gave his informed consent for the publication of this case report. The patient understands that his identity will not be revealed.

Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.amsu.2018.08.007>.

References

- [1] Lipa Bodneret, et al., Cutaneous sinus tract of dental origin in Children - a report of 28 new cases, *Pediatr. Dermatol.* 29 (4) (2012 Jul-Aug) 421–425.
- [2] P. Shaqil, B. Duane, K. Habib, Diagnostic challenges of cutaneous draining sinus tracts of odontogenic origin: a case report, *Dermatol. Surg.* 37 (2011) 1525–1527.
- [3] J.L. Cantatore, P.A. Klein, L.M. Lieblich, Cutaneous dental sinus tract, a common misdiagnosis: a case report and review of the literature, *Cutis* 70 (2002) 264–267 & #8232;.
- [4] M. Gorsky, I. Kaffe, A. Tamse, A draining sinus tract of the chin: report of a case, *Oral Surg. Oral Med. Oral Pathol.* 46 (1978) 583–587.
- [5] K.L. Spear, P.J. Sheridan, H.O. Perry, Sinus tracts to the chin and jaw of dental origin, *J. Am. Acad. Dermatol.* 8 (1983) 486–492.
- [6] R.A. Maple, T.F. Eichel, Cutaneous odontogenic sinus attributed to a maxillary first molar, *Gen. Dent.* 41 (1993) 168–170.
- [7] C.E. Urbani, R. Tintinelli, Patent odontogenic sinus tract draining to the midline of the submental region: report of a case, *J. Dermatol. (Tokyo)* 23 (4) (1996) 284–286.
- [8] J.L. Held, M.J. Yunakov, R.J. Barber, M.E. Grossman, Cutaneous sinus of dental origin: a diagnosis requiring clinical and radiologic correlation, *Cutis; Cutan. Med. Pediatr.* 43 (1) (1989) 22–24.
- [9] T. Sato, et al., Rare case of external dental fistula of the submental region misdiagnosed as inverted follicular keratosis and thyroglossal duct cyst, *Int. J. Surg. Case Rep.* 16 (2015) 39–43.
- [10] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill for the SCARE Group, The SCARE Statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [11] L.B. Kaban, Draining skin lesions of dental origin: the path of spread of chronic odontogenic infection, *Plast. Reconstr. Surg.* 66 (1980) 711–717.
- [12] Ronald S. Brown, et al., Cutaneous sinus tracts (or emerging sinus tracts) of odontogenic origin: a report of 3 cases, *Clin Cosmet Invg Dent* (2010) 63–67.

- [13] D.J. Sheehan, B.J. Potter, L. Davis, Cutaneous draining sinus tract of odontogenic origin: unusual presentation of a challenging diagnosis, *South. Med. J.* 98 (2005) 250–252.
- [14] G.A. Gioffi, G.T. Terezhalmay, H.L. Parlette, Cutaneous draining sinus tract: an odontogenic etiology, *J. Am. Acad. Dermatol.* 14 (1986) 94–100.
- [15] J.W. Harrison, W.J. Larson, The epithelized oral sinus tract, *Oral Surg. Oral Med. Oral Pathol.* 42 (1976) 511–517.
- [16] E. Tidwell, J.D. Jenkins, C.D. Ellis, B. Hutson, R.A. Cederberg, Cutaneous odontogenic sinus tract to the chin: a case report, *Int. Endod. J.* 30 (1997) 352–355.
- [17] Y. Nakamura, K. Hirayama, M. Hossain, K. Matsumoto, A case of an odontogenic cutaneous sinus tract, *Int. Endod. J.* 32 (1999) 328–331.
- [18] N. Mittal, P. Gupta, Management of extra oral sinus cases: a clinical dilemma, *J. Endod.* 30 (2004) 541–547.