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PRIKAZ SLUČAJA
CASE REPORT

Kavitarani Bhimappa Rudagi[1], Bhimappa Mallappa Rudagi[2]

Kožna fistula odontogenog podrijetla – pogrešno dijagnosticirana lezija: Prikaz dvaju slučajeva

Cutaneous Sinus Tract of Odontogenic Origin – A Misdiagnosed Lesion: Report of two cases

[1] Odsjek za restorativnu stomatologiju i endodonciju Stomatološkog fakuleta Sharad Pawar, Sawangi (meghe), Indija
Dept of Conservative Dentistry and Endodontics, Sharad Pawar Dental College, Sawangi (meghe), India
[2] Odsjek za oralnu i maksilofacijalnu kirurgiju Stomatološkog fakulteta Sharad Pawar, Sawangi (meghe), Indija
Dept of Oral and Maxillofacial Surgery Sharad Pawar Dental College, Sawangi (meghe), India

Sažetak
Svrha je ovog članka potaknuti raspravu o dvama slučajevima ekstraoralne fistule odontogenog podrijetla s naglaskom na dijagnosticiranje i liječenje. Inače, ekstraoralna kožna fistula rijetka je, ali dobro dokumentirana u literaturi. Vrlo se često pogrešno dijagnosticira kao lokalna kožna lezija, pa se i pogrešno liječi multiplim kirurškim ekscizijama i/ili sistemskim antibioticima, no ponovno se pojavljuje. To se događa zato što se zanemaruje primarna etiologija odontogenog podrijetla. U oba opisana slučaja pacijenti su naveli da pate od bolova i da imaju gnojni/krvavi iscjedak iz lezije. Klinički pregled i radiološka obrada pokazali su zahvaćene zube s periapikalnom lezijom. Nekirurški endodontski tretman bio je metoda izbora u prvom slučaju. U drugom je bila prijeko potrebna kirurška intervencija. Oba pacijenta reagirala su povoljno na terapiju i nisu imali nuspojave. Razumijevanje patogeneze kožnih fistula nastalih od odontogenih infekcija pomoći će da se što ranije postavi dijagnoza i omogućii uspješno liječenje.

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Adresa za dopisivanje
Dr Kavitarani Rudagi
Senior Lecturer
Dept. of Conservative Dentistry and Endodontics
Sharad Pawar Dental College
Sawangi (meghe).
District: Wardha
State: Maharashtra
tel 91 – 7709255022
kavitarudagi_11@rediffmail.com

Ključne riječi
propolis; stomatitis; alergeni; oralne lezije

Uvod

Kožna fistula odontogenog podrijetla rijetka je i često se teško dijagnosticira. Uglavnom je povezana fokusom preko kanala iz kojeg se upala drenira u područje lica ili vrata (1,2). Fistule češće nastaju nakon upala mandibularnih (80%) zuba negoli maksilarnih (20%) (3).

Klinički izgled ekstraoralne dentalne fistule može nalikovati na prištić, čir, nodul, te granulomatoznu i induriranu cističnu leziju. Kako su fistule klinički slične ostalim kožnim lezijama i razmjerno rijetke, često se zanemaruje dentalna etiologija te pacijente velikim dozama antibiotika liječe dermatolozi ili kirurzi, određuju im se ekscizije, biopsije, pa ponekad i zračenja. Pogrešna dijagnoza i kronični tijek lezije često loše utječu na estetiku lica jer pogrešno liječenje rezultira ožiljkastim tkivom i deformacijom kože (1 – 8).

Kožne fistule odontogenog podrijetla obično nastaju kao posljedica bakterijske infekcije pulpe kroz karioznu leziju ili traumu (9). Ako pacijent nije liječen u ranoj fazi, pulpa zuba nekrotizira i infekcija se širi iz korijenskog kanala u periradikularno područje te nastaje apikalni parodontitis. Poprat-

Introduction

Cutaneous sinus tract of dental origin is rare and often presents a diagnostic challenge. Cutaneous dental sinus tract is a duct which leads from a dental focus of infection to drain on to the face or neck (1, 2). These tracts tend to occur more frequently from infected mandibular teeth (80%) than maxillary teeth (20%) (3).

Clinically, a cutaneous dental sinus tract may resemble a pimple, ulcer, nodule, granulomatous lesion or indurated cystic area. These tracts have a similar clinical appearance to other facial lesions and are relatively uncommon, as the lesion develops, it is usually disregarded to be of dental origin, and patient requires treatment by a dermatologist or general surgeon and often undergoes multiple antibiotic regime, surgical excisions, biopsies and even radiotherapy. Misdiagnosis adds to the chronicity of the lesion and has profound effect on facial esthetics due to unnecessary treatment resulting in cutaneous scarring and dimpling. (1-8).

These cutaneous sinus tracts usually arise as a sequel of bacterial invasion of the dental pulp by a carious lesion or trauma

ni gnojni proizvodi infekcije traže mjesto gdje je u kosti i u mekom tkivu najmanji otpor (10). Te lezije često se pogrešno dijagnosticiraju i tretiraju. Ako se dentalni fokus ne liječi, pojavljuje se recidiv fistule (3, 8, 11).

U ovom članku opisali smo dva slučaja ekstraoralne fistule odontogenog podrijetla te njihovu dijagnostiku i liječenje.

Prvi slučaj

Zdrav muškarac u dobi od 22 godine primljen je u Zavod za restorativnu stomatologiju i endodonciju zbog izrasline ispod donje usne (slika 1. a, b). Žalio se na povremene bolove i iscjedak. U anamnezi je naveo dotadašnje preglede kod dermatologa i dvogodišnje liječenje kod tog specijalista. No unatoč tome lezija se nije povukla. Pacijent je u anamnezi naveo i dva pokušaja ekscizije.

Klinički pregled otkrio je izraslinu ispod donje usne promjera 0,3 centimetra i purulentni iscjedak na pritisak. Perkutorni test i test vitaliteta obavljeni su na zubima donje fronte. Zub 32 bio je blago osjetljiv na perkusiju. Zubi 32, 31 i 41 nisu reagirali na električni i termički test vitaliteta. Radiološka analiza otkrila je široku radiolucenciju povezanu sa zubom 32, te obliterirani korijenski kanal zuba 31 (slika 1. c). Nakon detaljne kliničke i radiološke obrade dijagnosticiran je kronični gnojni periapikalni parodontitis povezan sa zubima 31, 32 i 41.

Nekirurški endodontski tretman počeo je na zubima 31, 32 i 41. Pri instrumentaciji kanala korištena je Crown-Downova tehnika uz irigaciju 3-postotnim natrijevim hipokloritom (Hyposept, UPS Hygienes pvt Ltd). Kalcijev hidroksid (Ultracal XS; Ultradent, South Jordan, UT) rabio se kao medikamentni uložak između posjeta. Ekstraoralna fistula nestala je dva tjedna nakon početka liječenja korijenskih kanala (slika 2. d, e). Tri tjedna poslije inicijalne instrumentacije kanala, trajno su napunjeni štapićima gutaperke (Dentsply Maillefer, Ballaigues, Švicarska) i punilom AH-plus (Dentsply DeTrey GmbH, Nejmačka). Kontrolna radiološka snimka nakon godine dana pokazala je dobro cijeljenje periapikalne lezije i znatno poravnanje zuba 32 (slika 2. f).

Drugi slučaj

Zdrav šesnaestogodišnji mladić primljen je u Zavod za restorativnu stomatologiju i endodonciju jer se već 18 mjeseci žalio na kožnu leziju u submentalnoj regiji. Iz stomatološke anamneze bilo je jasno da je bio na nekoliko kirurških ekscizija, no lezija se vraćala. Razlog za neuspjeh takvog liječenja jest u činjenici da zubi nisu bili endodontski tretirani. Pacijent je naveo povremene bolove i iscjedak iz lezije.

Ekstraoralnim pregledom ustanovljena je kožna lezija dimenzija 2 x 2 centimetra u submentalnoj regiji (slika 2. a, b).

(9). If the treatment is not initiated at this stage, the pulp undergoes necrosis and infection spreads beyond the confines of the tooth in to the periradicular area resulting in apical periodontitis. The purulent by-products will seek the path of least resistance, when exiting from the root apex area and travelling through bone and soft tissues (10). These lesions are often misdiagnosed and are treated incorrectly. Unless the dental focus of infection is treated, recurrence is likely (3, 8, 11).

We have described two cases of cutaneous sinus tracts of odontogenic origin, their diagnoses and treatment.

Case 1

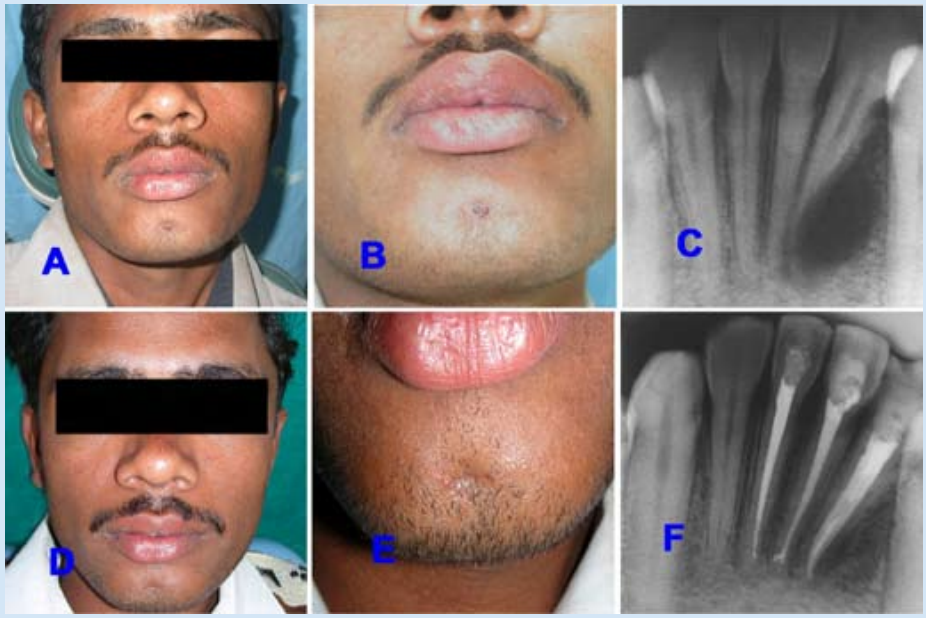
A healthy 22-year-old male patient came to the Department of conservative dentistry and endodontics with a nodule below the lower lip (Figure 1 A, B). He had intermittent pain and drainage through the lesion occasionally. Previous history of the patient revealed that he had consulted a dermatologist for the same problem and was treated for two years. The lesion recurred in spite of the repeated treatment regimen. He also reported a history of surgical excision twice for the same lesion.

Clinical examination revealed a nodule below the lower lip measuring 0.3cm in diameter and eliciting purulent discharge on palpation. Tooth percussion and vitality tests were carried out with lower anterior teeth. Tooth 32 was slightly tender on percussion. Teeth 32, 31, 41 failed to respond to electrical and thermal pulp tests. Radiographic examination revealed a broad radiolucency associated with 32 and obliteration of root canal space with 31(Figure 1 C). Chronic suppurative periapical periodontitis with an extraoral sinus tract was diagnosed in relation to 31, 32, and 41 after complete clinical and radiological examination.

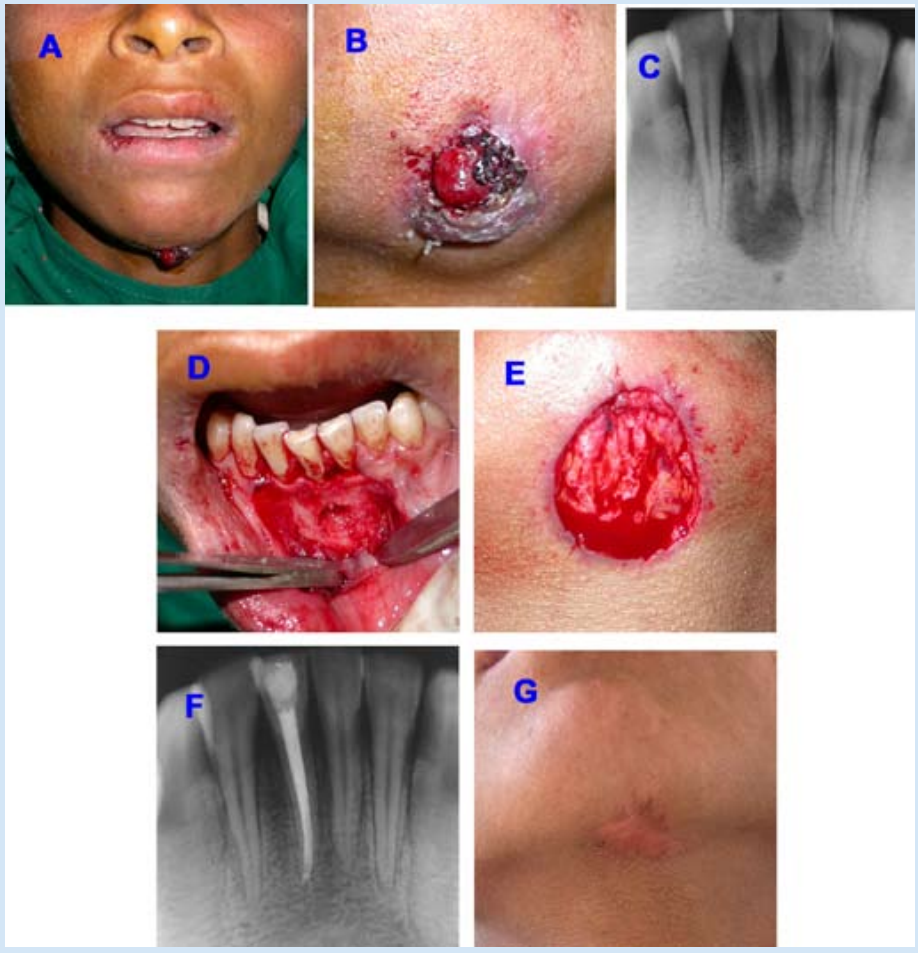
A non surgical endodontic treatment was started on teeth 31, 32, 41. The root canal system was cleaned and shaped using Crown-Down technique and irrigated with 3% sodium hypochlorite (Hyposept, UPS Hygienes pvt Ltd) as a disinfectant. Calcium hydroxide (Ultracal XS; Ultradent, South Jordan, UT) was used as an intracanal medication. The extraoral nodule disappeared in two weeks after the institution of root canal treatment (Figure 2 D, E). Three weeks after the initial presentation, the root canals were obturated with gutta-percha (Dentsply Maillefer, Ballaigues, Switzerland) and AH Plus sealer (Dentsply DeTrey GmbH, Germany). One year after, periapical radiographs showed signs of healing of periapical lesion with considerable amount of realignment of drifted tooth 32 (Figure 2 F).

Case 2

A healthy 16-year-old male patient was referred to the Department of conservative dentistry and endodontics with a draining cutaneous lesion in the submental region present for one and half year. The past dental history revealed that the patient had undergone multiple surgical excisions for the same lesion, but the cutaneous lesion always recurred. Previous surgical treatment had been unsuccessful because no dental treatment was done and only the cutaneous lesion was excised. He reported intermittent pain and drainage through the lesion.



Slika 1. a,b: Predoperativne slike; c: Predoperativna rtg. snimka; d,e: Postoperativna rtg. snimka; f: Kontrolna rtg. snimka nakon jedne godine
Figure 1 A, B: Pre-Operative photograph; C: Pre-Operative radiograph; D, E: Post-Operative photograph; F: Recall radiograph 1 year postoperatively



Slika 2. a,b: Predoperativna slika; c: Predoperativna rtg. snimka; d: Enukleacija ciste; e: Ekscizija ekstraoralne kožne lezije; f: Kontrolna rtg. snimka nakon jedne godine; g: Kontrolna slika nakon jedne godine
Figure 2 A, B: Pre-Operative photograph; C: Pre-Operative radiograph; D : Cyst enucleation; E: Excision of extra-oral cutaneous lesion; F: Recall radiograph 1 year postoperatively; G: Post-operative photograph at 1 year.

Oteklina i okolno tkivo bili su tvrdi i bolni na dodir. Pojavljivao se i gnojni iscjedak. Obavljen je rutinski intraoralni pregled, uključujući testove perkusije i vitaliteta donje fronte. Pritom je zub 41 reagirao negativno na test vitaliteta. Radiološkim pregledom ustanovljena je zaobljena radiolucencija povezana s korijenom zuba 41, a sličila je radikularnoj cisti (slika 2. c). Na osnovi kliničkih pregleda dijagnosticirana je radikularna cista uzrokovana nekrozom pulpe zuba 41 i ekstraoralna fistula.

Endodontski tretman počeo je na zubu 41 i to Crown-Downovom tehnikom uz irigaciju 3-postotnim natrijevim hipokloritom (Hyposept, UPS Hygienes pvt Ltd). Kalcijev hidroksid (Ultracal XS; Ultradent, South Jordan, UT) primijenio se kao intrakanalni uložak. Lezija nije pokazivala znakove cijeljenja. Iz nje je još povremeno curilo i pacijent se žalio na bolove četiri tjedna nakon endodontskog zahvata. Uzimajući u obzir sve čimbenike, odlučeno je da se obavi kirurška ekscizija lezije.

Prvi potez u cjelokupnoj terapiji jest završetak endodontske terapije. Punjenje korijenskog kanala obavljeno je štapićima gutaperke (Dentsply Maillefer, Ballaigues, Švicarska) i punilom AH-plus (Dentsply DeTrey GmbH, Njemačka) koristeći se tehnikom lateralne kondenzacije.

Slijedile su enukleacija ciste (periapikalna kirurgija) (slika 2. d) i ekscizija ekstraoralne fistule (slika 2. e). Metoda estetskog zbrinjavanja operacijske rane bila bi presađivanje kože na mjesto lezije, ali za taj zahvat pacijent nije potpisao pristanak. Na mjestu ekstraoralne lezije nastalo je ožiljkasto tkivo. Uzorak periapikalnog tkiva poslan je na PHD-analizu i tada je dijagnosticirana odontogena keratocista. Radiološka analiza periapeksa nakon godine dana pokazala je znakove cijeljenja periapikalne lezije.

Rasprava

Ekstraoralna fistula odontogenog podrijetla rijetka je, ali u literaturi dobro dokumentirana dijagnoza.

Kronična periapikalna infekcija može prouzročiti asimptomatični sinusni trakt koji obično rezultira vestibularnom fistulom u blizini zahvaćenog zuba. Ponekad se fistula vidi i na licu, što uglavnom ovisi o hvatištu mišića lica i okolnih tkiva u odnosu prema fokusu infekcije. Fistule se pojavljuju na licu ako je fokus infekcije superiorno od hvatišta mišića u maksili, odnosno inferiorno od hvatišta mišića u mandibuli. Apsces pronalazi mjesto najmanjeg otpora uz tkivne fascije dok ne izbije na koži (9, 12).

Pacijenti s ekstraoralnom fistulom odontogenog podrijetla obično nemaju zubobolju zato što se purulentni sadržaj drenira kroz fistulu te se smanjuje pritisak. Upravo se zbog toga obično zanemaruje njezina odontogena etiologija (1, 13).

Extraoral examination revealed a cutaneous lesion approximately 2x2cm in the submental region (Figure 2 A, B). The lesion and surrounding area were tender on palpation eliciting a purulent discharge and fixation of the lesion to the underlying tissue. Intraoral examinations consisting of routine clinical tests were conducted including tooth percussion and vitality testing of lower anterior teeth. Tooth 41 gave a negative response to vitality testing. Radiographic examination revealed a well circumscribed radiolucency associated with tooth 41 giving a radiographic impression of a radicular cyst (Figure 2 C). Based on clinical and radiographic findings, a diagnosis of a radicular cyst caused by pulpal necrosis with 41 associated with cutaneous sinus was made.

Endodontic treatment started with tooth 41, the root canal system was cleaned and shaped using Crown Down technique and irrigated with 3% sodium hypochlorite (Hyposept, UPS Hygienes pvt Ltd) as a disinfectant. Calcium Hydroxide (Ultracal XS; Ultradent, South Jordan, UT) was used as an intracanal medication. The lesion showed no signs of healing. There was an intermittent discharge still present and the patient complained of pain even after four weeks of root canal therapy. Hence the decision to perform a periapical surgery with excision of the extraoral lesion was made.

The first step of the treatment plan was to complete root canal therapy. Obturation was completed with gutta percha (Dentsply Maillefer, Ballaigues, Switzerland) and AH Plus sealer (Dentsply DeTrey GmbH, Germany) by lateral condensation technique.

The second step of treatment was periapical surgery (cyst enucleation) (Figure 2 D) and excision of the sinus tract and extraoral granulomatous lesion (Figure 2 E). Skin grafting for immediate esthetic repair was the treatment of choice; however, the patient's consent could not be obtained. The periapical tissue specimen was sent for histopathological examination. The periapical cyst was diagnosed histologically as an odontogenic keratocyst. The postoperative period was uneventful. One year later, periapical radiograph showed signs of healing of the periapical lesion.

Discussion

The cutaneous sinus tract of dental origin is an uncommon but well documented condition in the literature.

Chronic periapical infection around a tooth can produce a burrowing, and practically asymptomatic sinus tract that eventually drains within the mouth, commonly in the vestibule, adjacent to the affected tooth. However, it may drain on to the face, depending on the relationship of the muscle attachments and tissue planes of the face to the focus of infection. Sinus tracts form on the face if the focus of infection is superior to the muscle attachment in the maxilla or inferior to the muscle attachment in the mandible. An abscess travels via the route of least resistance along facial planes until exiting on the skin (9, 12).

The patients with cutaneous facial sinus tracts of dental origin often do not have obvious dental symptoms, as they provide an outlet for inflammatory exudates, allowing de-

Klinički pregled kožne fistule mora početi detaljnim anamnestičkim pristupom i kliničar mora biti svjestan da svaka fistula na licu ili vratu može biti odontogena. Ključne stavke svakog pregleda ekstraoralne fistule su vanjski izgled lezije i palpatorni nalaz sinusnog trakta povezanog s fistulom i podliježućom kosti (1). Intraoralni pregled može otkriti kariozne zube ili zube promijenjene boje. Ako su zahvaćeni, reagiraju negativno na test vitaliteta pulpe. Insercija štapića gutaperke u sinusni trakt te rendgenska slika obično pomažu pri određivanju izvora fokusa.

Diferencijalna dijagnoza uključuje prištić, aktinomikozu, orokutanu fistulu, ductus tyroglosus, fistulu žlijezde slinovnice, kroničnu tuberkulozu, gumu ili tercijarni sifilis, gnojni limfadenitis te malignitet (4, 8).

Nekirurška endodontska terapija (slučaj 1.) jest terapija izbora ako se zub može izliječiti (1). Kirurški zahvat (slučaj 2.) izbor je u slučaju da endodontski tretman na uspije. Winstock preporučuje tijekom terapije kiruršku eksciziju kožne fistule i sinusnog trakta te imedijatnu plastičnu rekonstrukciju kožne lezije (14, 15). Ako se zub ne može izliječiti, indicirane su ekstrakcija i kiretaža (1).

Sistemski antibiotici nisu bili potrebni jer je lezija lokalizirana. Liječenje antibioticima obično je neuspješno zato što uglavnom rezultira privremenim nestankom gnojnog iscjetka (7, 16). Preporučuje se jedino ako je potrebna kirurška intervencija. Plastično-kirurški zahvat potreban je ako tijekom cijeljenja nakon ekscizije kožne lezije nastaje ožiljkasto tkivo (1, 14, 17).

Histološki se te lezije sastoje od granulomatoznog ili vezivnog tkiva infiltriranog upalnim stanicama (9, 17, 18).

Zaključak i klinička primjena

Kronične odontogene upale jedan su od najčešćih uzroka fistula lica i vrata.

U medicinskoj literaturi navodi se da su mnogi pacijenti s dijagnozom ekstraoralne fistule odontogenog podrijetla bili podvrgnuti neuspješnim pokušajima incizije i drenaže, kurama antibiotika (3, 5, 6, 8), zračenjima (5), kauterizaciji (5), intralezijskim injekcijama steroida (19), oralnim steroidima (19) te laserskim terapijama (19).

Obiteljski liječnik treba poznavati nesvakidašnje simptome periapikalnih infekcija. Razumijevanje patogeneze ekstraoralne fistule odontogenog podrijetla i detaljna stomatološka anamneza pomažu da se postavi točna dijagnoza i odredi liječenje bez nepotrebnih medicinskih zahvata.

compression and therefore, possible dental etiology may be overlooked (1, 13).

Evaluation of a cutaneous sinus tract must begin with a thorough history and the clinician's awareness of the fact that any cutaneous lesion of the face and neck can be of dental origin. Key points for extraoral clinical examination are the gross appearance of the lesion and palpation of cordlike tract attached to the underlying bone (1). Intraoral examination may reveal carious or discolored teeth. The involved teeth respond negatively to pulp vitality tests. Periapical radiograph taken by a gutta-percha point inserted in the sinus is helpful in tracking the origin of the lesion.

Differential diagnosis includes pustule, actinomycosis, orocutaneous fistula, thyroglossal duct cyst, salivary gland fistula, chronic tuberculosis, gumma of tertiary syphilis, suppurative lymphadenitis and malignancy (4, 8).

Non surgical endodontic therapy (as in Case 1) is the treatment of choice if the tooth is restorable (1), surgical endodontic therapy (as in Case 2) can also be considered if nonsurgical endodontic treatment fails. Winstock recommends excision of the cutaneous lesion and stoma in continuity at the time of treatment of the dental pathology with immediate plastic repair of the cutaneous site (14, 15). If the tooth is nonrestorable, extraction and curettage will be indicated (1).

No systemic antibiotics are required since the lesion is a localized entity. The antibiotic therapy is actually unsuccessful and may be misleading because the drainage may stop temporarily (7, 16). If surgical intervention is sought, then antibiotic coverage is justified. Plastic surgery repair may be needed at a later stage if the healing results in cutaneous retraction (1,14,17).

Histologically, these lesions consist of granulation tissue or connective tissue heavily infiltrated with inflammatory cells (9, 17, 18).

Conclusion and Clinical Significance

Chronic draining dental infection is one of the most common causes of sinus tracts of the face and neck.

In the medical literature, approximately half the patients with cutaneous sinus tract of dental origin have undergone multiple unsuccessful attempts at incision and drainage, numerous antibiotic therapies (3, 5, 6, 8), radiation therapy (5), electrodessication(5), intralesion injection of steroid (19), oral steroid(19) and laser therapy(19).

Physicians should remain aware of the unusual presentation of periapical infection. An understanding of the pathogenesis of cutaneous fistulae arising from dental infection coupled with a thorough history will lead to accurate early diagnosis and treatment, without unnecessary and unsuccessful treatments.

Abstract
The aim of the present article was to discuss two different case reports of the extra-oral odontogenic cutaneous sinus tracts with emphasis on their diagnoses and treatment plans. Odontogenic cutaneous sinus is a rare but well documented condition. It is usually misdiagnosed as a local skin lesion and mistreated by multiple surgical excisions and/or systemic antibiotics with eventual recurrence. This is because the primary dental etiology is overlooked. We have described two cases of extraoral cutaneous sinus tract of an odontogenic origin. In both cases there were complaints of pain and purulent/hemorrhagic discharge from the lesions. Clinical and radiological examination showed inactive teeth, which were infected periapically in both cases. Non-surgical endodontic treatment was successful in Case 1. Surgical intervention was performed in Case 2. Both patients responded well and healing was uneventful. An understanding of the pathogenesis of cutaneous sinus tract arising from dental infection will lead to proper early diagnosis and successful treatment.

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Address for correspondence
Dr Kavitarani Rudagi, Senior Lecturer
Sharad Pawar Dental College
Dept of Conservative Dentistry and Endodontics
Sawangi (meghe).
District: Wardha
State: Maharashtra
Phone No: 91-7709255022
kavitarudagi_11@rediffmail.com

Key words
Propolis; Stomatitis; Allergens; Oral Lesions

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