

# Extraoral Sinus Tracts of Odontogenic Origin: A Case Series

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Article Info	ABSTRACT
<i>Article type:</i> Case Report	Extraoral sinus tracts of odontogenic origin often develop as the result of misdiagnosis of persistent dental infections due to trauma, caries, or periodontal disease. Due to these lesions' imitation from cutaneous lesions, misdiagnosis, and mismanagement, which we frequently encounter, this article aims to describe four cases with manifestations in different parts of the face and the neck. Patients were referred to an endodontist with a history of several surgical procedures and/or antibiotic therapy due to misdiagnosis. After comprehensive examinations, root canal treatment was performed. The resolution of signs and symptoms during the follow-up period confirmed the correct diagnosis. Dermatologists and other physicians should be aware of the possibility of the relationship of extraoral sinus tracts with dental infections. Precise examination and taking a comprehensive history can aid to prevent unnecessary and incorrect therapeutic and/or pharmaceutical interventions. Elimination of dental infection leads to complete
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Email: m69_saber@yahoo.com	recovery in such patients. Keywords: Endodontics; Misdiagnosis; Infection; Odontogenic Cysts

#### INTRODUCTION

Apical periodontitis is an inflammatory disease that may occur in acute, subacute, or chronic forms. Chronic apical periodontitis may manifest in the form of a dental granuloma or periradicular cyst and is characterized by invasion to the connective tissue, destruction, and repair [1]. A sinus tract may form in some cases of chronic inflammation [2]. A sinus tract is defined as a communication pathway between a confined inflammatory site and an epithelial surface [3]. Depending on the path with the lowest resistance, the sinus tract opening may be located intraorally or extraorally. In the intraoral form, the sinus tract opening may be in the attached buccal mucosa or the vestibule. In the extraoral form, the sinus

tract may open anywhere on the face or the neck. It is commonly found on the cheeks, the chin, the angle of the mandible, and occasionally on the nasal floor [4-7]. Extraoral sinus tracts are often related to the mandibular teeth with a prevalence of 80-87% [8]. Extraoral sinus tracts of dental origin can be misdiagnosed with many conditions in differential diagnosis such as local cutaneous infections, ingrown hair or occluded sweat gland ducts. osteomyelitis, neoplasms. tuberculosis, actinomycosis, and congenital midline sinus of the upper lip [9-16]. The cutaneous sinus tract of odontogenic origin is relatively common and may be misdiagnosed due to the absence of dental symptoms, variable morphology, and uncommon location

[17]. Given the importance of the diagnosis and management of these lesions and avoiding unnecessary treatments, the case series reports four cases of misdiagnosed extraoral sinus tracts of odontogenic origin in different parts of the face and the neck.

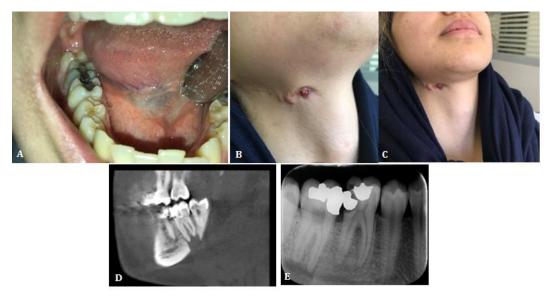
## CASE REPORT

#### Case 1:

A 27-year-old female referred, complaining of an extraoral sinus tract opening. The patient's medical history was unremarkable, and she was categorized as ASA I. The patient recalled the development of an extraoral sinus tract on the right submandibular region from six years ago. The patient visited an ear, nose, and throat (ENT) specialist upon noticing it. The specialist had prescribed chlorhexidine (CHX) mouthwash for 2 weeks. Due to no recovery and continuous pus discharge from the sinus tract, the patient referred to an infectious disease specialist, who prescribed daily use of 100 mg doxycycline for 6 months. The patient underwent sonography, which revealed a hypoechoic mass with unclear margins and no calcification. Due to the continuation of pus discharge, the patient was admitted to a hospital, and a general surgeon surgically resected the lesion.

However, the patient did not have the pathology

report of the lesion with her. Pus discharge and the symptoms were resolved after the surgical procedure. However, the symptoms recurred one year later. The patient presented again to the general surgeon, and this time, she was diagnosed with tuberculosis. Polymerase chain reaction and cell culture were performed, which were found to be negative for Mycobacterium tuberculosis. The patient was scheduled for another surgical procedure with a diagnosis of a branchial cyst. The surgical procedure was performed, and the pathology report indicated an infected wall of a sinus tract. Pus discharge continued for another year after surgery, and the patient was referred to a maxillofacial surgeon, who recommended another surgical procedure, which was rejected by the patient. The patient used herbal medications for one year but the pus discharge continued. The patient presented to our dental school. In the first visit, a fistula on the right side of the neck was noticed. The clinical and radiographic examinations were performed (Fig. 1 A-D). The right mandibular first and second molars gave a negative response to pulp vitality tests. Teeth #30 and #31 were not sensitive to percussion and had no mobility. Periapical radiographs revealed severe bone destruction in the periapical region of tooth #31 and periodontal ligament (PDL) widening around the apex of tooth #30.



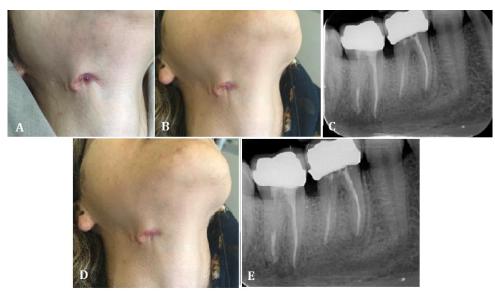
**Fig. 1.** (A, B, C) Photographs of the patient in the first visit. (D) Periapical radiographs of teeth #30 and #31. (E) Sagittal view of cone-beam computed tomography

Cone-beam computed tomography (CBCT) was then requested for the patient, which indicated extensive bone destruction in the body of the mandible (Fig. 1E). Teeth #30 and #31 underwent root canal treatment (RCT). In the first treatment session, local anesthesia was induced by the injection of 2% lidocaine and 1:80,000 epinephrine (Persocaine-E, Daru Pakhsh, Tehran, Iran), and an access cavity was prepared in tooth #31. The working length was then determined. The canals were cleaned using the Edge Taper Platinum rotary system (EdgeEndo, Albuquerque, NM, USA) to #F2 file along with irrigation with 5.25% sodium hypochlorite (NaOCl; Hypo-EndoX, Morvabon, Iran). A mixture of calcium hydroxide powder (Golchadent, Tehran, Iran) and 2% CHX (MORVA-Sept, Morvabon, Tehran, Iran) was applied to the canals as an intracanal medicament. The second session was scheduled one week later. In the second session, there was no pus discharge, and the sinus tract was closed (Fig. 2A). Thus, RCT was completed, and the canals were filled with gutta-percha and AH-26 sealer (Dentsply, Konstanz, Germany) using the cold lateral compaction technique. Tooth #30 also underwent RCT, and the patient was referred to a prosthodontist for the final restoration of the respective teeth. The first follow-up session was scheduled at 3 months after the final restoration of the teeth.

At the 3- and 6-month follow-ups, the patient was asymptomatic clinically and radiographically, and the periapical radiograph showed evidence of bone regeneration (Fig. 2B-E).

#### Case 2

Our second case was a 16-year-old male complaining of puss discharge from an opening in the submental region. The patient's medical history confirmed that he was ASA I. The extraoral examinations revealed no lymphadenopathy, tenderness, or facial asymmetry. The patient recalled trauma to the mandibular incisors 2 years earlier, which led to a crown fracture of tooth #25. The patient had visited a general dentist and received antibiotics with no emergency dental treatment. He mentioned the development of an extraoral sinus tract in the submental region one year earlier with episodes of pus discharge. The patient referred to an oral medicine specialist for the assessment of the pulp vitality status of his anterior teeth, and then, was referred to the Endodontics Department of Tehran University of Medical Sciences. The extraoral examinations revealed that the sinus tract was closed and had no active pus discharge. The intraoral examination revealed that the patient had good oral hygiene. The incisal edge of tooth #25 had chipping but the tooth was not mobile. The probing depth was within the normal range of 2-3 mm.



**Fig. 2.** (A) Photograph of the patient 1 week after treatment, (B) Photograph and (C) Radiograph of the patient 3 months after treatment, (D) Photograph and (E) Radiograph of the patient at the 6-month follow-up



**Fig. 3.** (A, B) Preoperative photographs, (C) radiograph, (D) inter-operative photograph and (E) second-visit radiograph after obturation

The tooth responded negatively to pulp vitality tests but was slightly sensitive to percussion. The radiographic examination revealed a radiolucent lesion with ill-defined borders in the periapical region of tooth #25 (Fig. 3A-C). The diagnosis of pulp necrosis following coronal fracture due to trauma and a chronic apical abscess was made. Local anesthesia was induced by the injection of 2% lidocaine and 1:80,000 epinephrine. An access cavity was prepared, and isolation was performed with clamps and a rubber dam. The working length was determined, and the root canal was instrumented using the Edge Taper Platinum rotary system. The canal had no pus discharge. A mixture of calcium hydroxide and 2% CHX was applied as an intracanal medicament. The second treatment session was scheduled 2 weeks later. The extraoral sinus tract had healed (Fig. 3 D). Thus, the root canal was filled with gutta-percha and AH-26 sealer with the cold lateral compaction technique (Fig. 3 E).

At the 2-month follow-up session (Fig. 4 A, B), the patient had no complaint of pus discharge from the extraoral sinus tract, and the opening had been completely healed. The respective tooth was asymptomatic on intraoral clinical examination. Radiographics showed the initiation of trabeculation in the periapical region. At the 5-month recall session (Fig. 4 C, D), the extraoral sinus tract opening had completely healed, and the periapical lesion had shrunk on the radiographic examination. At the 1-year follow-up (Fig. 4 E, F), the respective tooth was asymptomatic, and the periapical lesion had completely healed.



**Fig. 4.** (A) Clinical and (B) radiographic views at the 2month follow-up (C, D) at the 5-month follow-up, and (E, F) at the 1-year follow-up



**Fig. 5.** (A) Computed tomography scan of the patient, (B) extraoral and (C) intraoral photographs of the patient in the first session, (D) periapical radiograph of the respective tooth in the first treatment session. (E) Extraoral photograph of the patient 1 week after first treatment session. (F) Periapical radiograph and (G) extraoral photograph of the patient at the 3-month follow-up

## Case 3:

The third case was a 31-year-old female patient complaining of a pimple on her right cheek. The patient was systemically healthy. She had visited a general practitioner earlier and had been referred to a surgeon. The surgeon had suggested an excisional biopsy. She had undergone a computed tomography scan (Fig. 5A).

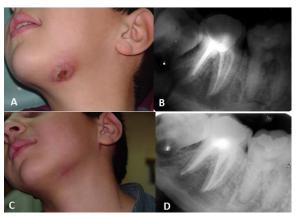
The radiologist's report described an abnormal density on the right side of the maxilla. Fortunately, the patient was advised by her radiologist to visit a dentist. The extraoral examination revealed a dense mass in the check area (Fig. 5B).

On the intraoral examination, her maxillary right first premolar had an incomplete previous RCT with an open access cavity without any temporary restoration (Fig. 5C). The radiographic examination revealed that tooth #5 had a small periapical radiolucency and PDL widening (Fig. 5D). RCT was suggested and started after obtaining written informed consent. In the first session, local anesthesia was induced by the injection of 2% lidocaine plus 1:80,000 epinephrine.

An access cavity was completed, and isolation was performed by a rubber dam and clamps. The root canals were irrigated with 2.5% NaOCl during root canal preparation. Then, the canals were rinsed with normal saline, and a mixture of calcium hydroxide and 2% CHX was applied as an intracanal medicament. The access cavity was temporarily restored with reinforced zinc oxide eugenol (ZOE) cement (Zonalin, Kem Dent, ADP, Swindon, UK). The second session was scheduled one week later. In the second treatment session, the significant healing of the sinus tract opening on the cheek was noticed (Fig. 5E). RCT was accomplished, and the root canals were filled with gutta-percha and AH-26 sealer by the cold lateral compaction technique. The patient was then referred to a restorative dentist for the final restoration of the tooth. The patient had no clinical or radiographic signs/symptoms at the 3-month follow-up (Fig. 5F, G).

## Case 4:

The fourth case was a 12-years-old boy complaining of a large ulcerative pimple right beneath the angle of the mandible on the left side. The patient was systemically healthy. The lesion had developed one year earlier, and the patient had used several ointments under the supervision of a dermatologist with no dermatologist finally improvement. His referred him to an endodontist. The intraoral and extraoral examinations were performed, and the left maxillary and mandibular teeth were thoroughly examined. Tooth #17 had undergone RCT, and the intraoral soft tissue was intact with no abnormality. Periapical radiography of the left mandibular quadrant was requested, which revealed that tooth #17 had an incomplete RCT and a radiolucent periapical lesion (Fig. 6A, B). The patient recalled RCT of this tooth 2 years earlier. The diagnosis of a chronic apical abscess was made based on the clinical and radiographic signs and symptoms, and the patient underwent nonsurgical endodontic retreatment. In the first treatment session, the conventional inferior alveolar nerve block was administered by the injection of 2% lidocaine and 1:80,000 epinephrine.



**Fig. 6.** (A) Photograph and (B) periapical radiograph of the patient in the first treatment session. (C) Photograph and (D) periapical radiograph of the patient at 2 weeks after treatment

The amalgam restoration was removed, isolation was performed with a rubber dam and clamps, and gutta-percha was removed using a chloroform solution (Merck, Darmstadt, Germany) and #25 H-file ((Micro Mega, Prodonta Sk Geneva, Switzerland). The root canals were rinsed with 5.25% NaOCl and instrumented by the ProTaper rotary system (Dentsply Maillefer, Baillagues, Switzerland) to #F2 file. After rinsing with saline, a mixture of calcium hydroxide and 2% CHX was used as an intracanal medicament, and reinforced ZOE cement was applied for temporary restoration of the access cavity. The second session was scheduled 2 weeks later, which revealed a closed sinus tract. RCT was accomplished, and the root canals were filled using guttapercha and AH-26 sealer with the cold lateral compaction technique (Fig. 6C, D). The patient was referred to a restorative dentist for the final restoration of the respective tooth. The patient did not show up for the next follow-ups.

#### DISCUSSION

Misdiagnosis and mismanagement of odontogenic extraoral sinus tracts have been previously reported in the literature [2,17-20]. One major reason for misdiagnosis is that these lesions may mimic some cutaneous lesions; therefore, at first, the patient often refers to a general surgeon or a dermatologist instead of a dentist, which often leads to mismanagement, unnecessary treatments such as topical and oral antibiotic therapy or surgical intervention, and different diagnostic tests [21].

Precise intraoral and extraoral examinations are imperative for the correct diagnosis and treatment planning. Dental practitioners should be careful with the interpretation of signs and symptoms such as deep dentin caries, inappropriate restorations, periodontal problems, and history of dental trauma [22].

The diagnosis of necrotic pulps in teeth can be challenging in the initial phases and may be enhanced by advanced diagnostic workup and radiographic detection of related bone loss [23]. A previous clinical study assessed periradicular inflammation and its relationship with the development of sinus tracts and showed that sinus tracts with endodontic origin commonly manifest intraorally. Extraoral sinus tracts are related to the mandibular teeth in 80% of the cases. and pus discharge often occurs through an opening in the chin or the submandibular region [24]. Additionally, in the current study, an uncommon case of an endodontic originated sinus tract in the neck area is presented. The cornerstone of treatment in such cases is the elimination of infection from the root canal system, which often leads to the healing of the extraoral sinus tract. For this purpose, NaOCl is often used for root canal irrigation during root canal instrumentation, and then, calcium hydroxide in combination with CHX is applied to the root canal system as an intracanal medicament until the resolution of the extraoral sinus tract [19]. Calcium hydroxide is commonly used as an intracanal medicament due to its potent antibacterial properties against the majority of endodontic microflora [25]. The high pH of calcium hydroxide formulation changes the biological properties of bacterial lipopolysaccharides in the cell wall of Gram-negative bacteria, inactivates the membrane transfer mechanisms, and leads to bacterial toxicity [26]. Antibacterial and bacteriostatic properties, enhancement of healing, high pH, which leads to subsequent stimulation of fibroblasts, inhibition of internal root resorption, neutralization of acidic pH, low cost, and easy application are among other advantages of calcium hydroxide [27]. CHX is a broad-spectrum antimicrobial agent, which is effective against Gram-positive and Gramnegative bacteria and yeasts. Due to its cationic property, it attaches to the tooth structure and hydroxyapatite, causing its optimal substantivity [28]. The duration of exposure and concentration of CHX affect the substantivity of its antibacterial activity. At low concentrations (0.005% to 0.01%), only one layer of CHX forms on the tooth surface. However, at higher concentrations (>0.02%), several layers of CHX form on the tooth surface and provide a reservoir that releases CHX as soon as its concentration drops in the surrounding environment [29]. One major drawback of calcium hydroxide is its inability to eliminate Enterococcus faecalis. Evidence shows that CHX can effectively eliminate the Enterococcus faecalis biofilm from the root canal system when used as an intracanal medicament [30,31]. Evidence shows that a combination of calcium hydroxide and CHX has optimal antimicrobial efficacy against Enterococcus faecalis [31,32]. In all of our reported cases, 2.5%-5.25% NaOCl was used as an intracanal irrigating solution, and a combination of calcium hydroxide and 2% CHX was used as an intracanal medicament until the healing of the extraoral sinus tract. If the origin of an extraoral sinus tract is correctly recognized, it is expected to heal and disappear within 7 to 14 days [6]. Follow-up examinations in all cases revealed clinical healing of the sinus tract, and periapical radiography showed healing of the periapical lesion and alleviation of signs and symptoms. However, our first case had a scar due to previous surgical procedures at the site. Cosmetic surgical procedures or laser therapy can be suggested for such patients.

#### CONCLUSSION

Root canal therapy by using antimicrobial irrigants and intracanal medicaments is the treatment of choice in these instances. At first, no surgical intervention is recommended.

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# **CONFLICT OF INTEREST STATEMENT** None declared.

#### REFERENCES

1. Abbott PV. Classification, diagnosis and clinical manifestations of apical periodontitis. Endod Top. 2004 Jul;8(1):36-54.

2. Curvers F, De Haes P, Lambrechts P. Nonsurgical endodontic therapy as treatment of choice for a misdiagnosed recurring extraoral sinus tract. Eur Endod J. 2017 Jan 1;2(1):13.

3. Glossary: Contemporary Terminology for Endodontics. American Association of Endodontists, 1994. Available at: https://www.endoexperience.com/documents/0 3GlossaryofEndoTerms\_Web.pdf/ Accessed June 23, 2019.

4. Heling I, Rotstein I. A persistent oronasal sinus tract of endodontic origin. J Endod. 1989 Mar 1;15(3):132-4.

5. Gupta M, Das D, Kapur R, Sibal N. A clinical predicament—diagnosis and differential diagnosis of cutaneous facial sinus tracts of dental origin: a series of case reports. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2011 Dec 1;112(6):e132-6.

6. Johnson BR, Remeikis NA, Van Cura JE. Diagnosis and treatment of cutaneous facial sinus tracts of dental origin. J Am Dent Assoc. 1999 Jun;130(6):832-6.

7. Mittal N, Gupta P. Management of extra oral sinus cases: a clinical dilemma. J Endod. 2004 Jul 1;30(7):541-7.

8. Guevara-Gutiérrez E, Riera-Leal L, Gómez-Martínez M, Amezcua-Rosas G, Chávez-Vaca CL, Tlacuilo-Parra A. Odontogenic cutaneous fistulas: clinical and epidemiologic characteristics of 75 cases. Int J Dermatol. 2015 Jan;54(1):50-5.

9. Katou F, Motegi K. Congenital midline sinus of the upper lip: Report of a case. Int J Oral Maxillofac Surg. 1989 Aug 1;18(4):237-8.

10. Craig RM, Andrews JD, Wescott WB. Draining fistulas associated with an endodontically treated tooth. J Am Dent Assoc. 1984 May;108(5):851-2.

11. Goldstein BH, Sciubba JJ, Laskin DM. Actinomycosis of the maxilla: review of literature and report of case. J Oral Surg. 1972 May;30(5):362-6.

12. Purohit SD, Mathur BB, Gupta PR, Agarwal KC, Hathi HH. Tuberculous fistula of cheek: Report of a case. Oral Surg Oral Med Oral Pathol. 1985 Jul 1;60(1):41-2.

13. Mahler D, Joachims HZ, Sharon A. Cutaneous dental sinus imitating skin cancer. Br J Plast Surg. 1971 Jan 1;24:78-81.

14. Jacobs J, Shocket E. Dermal fistula of dental origin masquerading as a skin cancer: Report of a case. Oral Surg Oral Med Oral Pathol Oral Radiol. 1965 Feb 1;19(2):184-7.

15. Yasui H, Yamagushi M, Lchimiya M, Yoshikawa Y, Hamamoto Y, Muto M. A case of cutaneous odontogenic sinus. J Dermatol. 2005;32:852-5.

16. Cioffi GA, Terezhalmy GT, Parlette HL. Cutaneous draining sinus tract: an odontogenic etiology. J Am Acad Dermatol. 1986 Jan 1;14(1):94-100.

17. Lee EY, Kang JY, Kim KW, Choi KH, Yoon TY, Lee JY. Clinical characteristics of odontogenic cutaneous fistulas. Ann Dermatol. 2016 Aug 1;28(4):417-21.

18. Cohenca N, Karni S, Rotstein I. Extraoral sinus tract misdiagnosed as an endodontic lesion. J Endod. 2003 Dec 1;29(12):841-3.

19. Pasternak-Júnior B, Teixeira CS, Silva-Sousa YT, Sousa-Neto MD. Diagnosis and treatment of odontogenic cutaneous sinus tracts of endodontic origin: three case studies. Int Endod J. 2009 Mar;42(3):271-6.

20. Swales KL, Rudralingam M, Gandhi S. Extraoral cutaneous sinus tracts of dental origin in the paediatric patient. A report of three cases and a review of the literature. Int J Paediatr Dent. 2016 Sep;26(5):391-400.

21. Salamat K, Rezai RF. Nonsurgical treatment of extraoral lesions caused by necrotic nonvital tooth. Oral Surg Oral Med Oral Pathol. 1986 Jun 1;61(6):618-23.

22. Witherow H, Washan P, Blenkinsopp P. Midline odontogenic infections: a continuing diagnostic problem. Br J Plast Surg. 2003 Mar 1;56(2):173-5.

23. Soares JA, De Carvalho FB, Pappen FG, Araújo GS, De Pontes Lima RK, Rodrigues VM, et al. Conservative treatment of patients with periapical lesions associated with extraoral sinus tracts. Aust Endod J. 2007 Dec;33(3):131-5.

24. Gupta R, Hasselgren G. Prevalence of odontogenic sinus tracts in patients referred for endodontic therapy. J Endod. 2003 Dec 1;29(12):798-800.

25. Kar S, Bera S, Karmakar D. Comparative evaluation of the antibacterial efficacy of calcium hydroxide as an intracanal medicament and combination of calcium hydroxide with food preservatives and proton pump inhibitor against Enterococcus faecalis-an invitro study. Paripex Indian J Res. 2020 Jan 30;9(1). 10.36106/paripex/7300841.

 Pannu R, Berwal V. Calcium Hydroxide in Dentistry: A Review. J Appl Dent Med Sci. 2017;3:3.
 Agrawal V. Calcium hydroxide: A miracle munition. Indian J Dent Res Rev. 2011;1:16-8.

28. Hjeljord LG, Rölla G, Bonesvoll P. Chlorhexidine–protein interactions. J Periodont Res. 1973 Dec;8:11-6.

29. Emilson CG, Ericson T, Heyden G, Magnusson BC. Uptake of chlorhexidine to hydroxyapatite. J Periodont Res. 1973 Dec;8:17-21.
30. Hugo WB, Longworth AR. Some aspects of the mode of action of chlorhexidine. J Pharm Pharmacol. 1964 Oct;16(10):655-62.

31. Ghabraei S, Bolhari B, Sabbagh MM, Afshar MS. Comparison of Antimicrobial Effects of Triple Antibiotic Paste and Calcium Hydroxide Mixed with 2% Chlorhexidine as Intracanal Medicaments Against Enterococcus faecalis Biofilm. J Dent (Tehran). 2018 May;15(3):151-160.

32. Nag M, DivyaLalitha N, Manipal S, Bharathwaj VV, Prabu D. Chlorhexidine Loaded Calcium Hydroxide as a Potential Antimicrobial Intracanal Medicament–A Systematic Review. J Pharm Sci Res. 2019 Sep 1;11(9):3210-6.