

Case Report

Anterior Mandibular Lingual Salivary Gland Defect Mimicking an Odontogenic Cyst- A Case Report

Venkatesh Anehosur¹, Kiran Radder², Harish K³, Veda Hegde⁴

¹Professor and Head of the department, ²Associate Professor, ³Post-graduate student, Dept. of Oral and Maxillofacial surgery, ⁴Professor, Professor, Dept. of Oral & Mnaxillofacial Pathology, SDM Craniofacial Surgery and research centre, SDM College of Dental Sciences and Hospital, Sattur, Dharwad - 580 009

*Corresponding Author : Venkatesh Anehosur, Professor & Head, Department of Oral & Maxillofacial Surgery, SDM Craniofacial Surgery & Research Centre, SDM College of Dental Sciences & Hospital, Sattur, Dharwad - 580 009.

Received : 10.10.2017

Review Completed : 15.12.2017

Accepted : 16.12.2017

Abstract

Stafne's bone defects (SBD) are usually asymptomatic and appears as a radiolucent lingual/buccal bone lesions of the lower jaw and are frequently caused by soft tissue inclusion. SBDs bone defects are considered to be an anatomic condition and most often seen in the posterior part the mandible below the mandibular canal. This is a case of anterior mandibular SBD, which was accidentally detected in a patient with mandibular fracture.

Keywords : Stafne bone defect, parasymphysis fracture, submandibular gland, sublingual gland

Access this article online
Quick Response Code




Introduction

The Stafne bone cavity was first described by Edward C. Stafne in 1942[1]. Stafne's bone defects (SBD) are usually asymptomatic and appear as a radiolucent jaw lesions and are frequently caused by soft tissue inclusion. Other terms used to describe this entity, include Aberrant or Ectopic salivary gland, Static or Idiopathic defects or cavity, Mandibular salivary gland inclusions, Lingual mandibular bone cavity and Stafne cyst, defect or cavity[2,3].

SBDs bone defects are considered to be an anatomic rather than a pathological condition and most often seen in the posterior part the mandible below the mandibular canal. Anterior salivary gland inclusions are considered to be rare and often they found between or below the root apices of canine and premolars. Due their close contact with the root apices, they mimic periapical lesion [4,5]

The purpose of this case report is to describe a case of anterior lingual mandibular salivary gland defect, which was detected accidentally in a patient who had mandibular fracture. The lesion was radiographically presented as an odontogenic cyst.

Case report

A 52 years old male patient reported to our hospital SDM Craniofacial unit, with a complaint of pain in the right side of lower jaw since 7 days. He gave alleged history of RTA, 7 days back and injury to his right side of face with a diagnosis of right parasymphysis fracture of mandible. Fracture was confirmed with panoramic radiograph, which also revealed a well-defined cystic lesion in the left side of anterior mandible, just below the root apices involving the lateral incisor, canine and first premolar teeth with root resorption (Fig. 1). Lesion was provisionally diagnosed as an odontogenic cyst.

Lesion was asymptomatic and the patient was unaware of its presence. Pulp vitality testing in relation to involved teeth revealed normal response. CT scan of face revealed a well-defined uniform bony defect in the periapical region of left lateral incisor, canine and first premolar with complete loss of lingual cortical plate continuity (Fig. 2)

Patient was explained about need for ORIF of fracture segments along with enucleation of cystic lesion under general anaesthesia and written consent was taken. Fractured segments were reduced and fixed using miniplates. Cystic cavity was exposed after creating a window in the labial cortex. Cystic contents were enucleated. Specimen was sent for histopathological examination. Post-operative period was uneventful.

Histopathological study revealed that there was presence of lobules of predominantly mucous acini and ducts. Lobules of adipocytes between glandular tissue (Fig.3). The biopsy report suggestive of salivary glandular tissue with inflammatory infiltrate. So overall features suggestive of "Stafne bone defect".



Fig 1 : Pre op panoramic radiograph showing radiolucent lesion in the anterior mandible on left side and right para-symphysis fracture (red arrow)

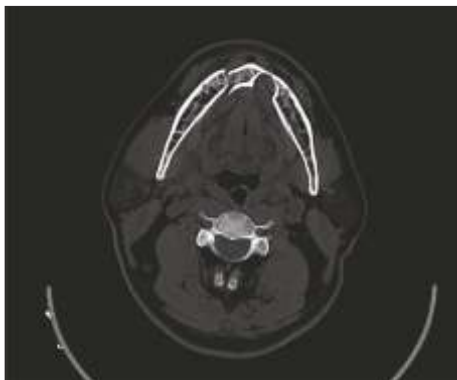


Fig 2: CT scan axial view showing lingual bony defect and fracture line.

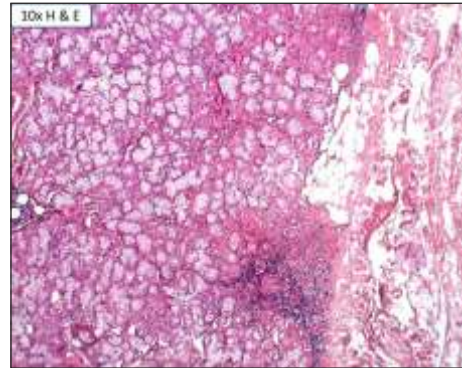


Fig 3 : Histopathology, 10x H & E stained section revealed, partially encapsulated salivary gland tissue with lobules of predominantly mucous acini.

Discussion

Stafne bone defect represents a focal concavity of the cortical bone on the lingual surface of the mandible. This is a developmental defect contains portion of submandibular or sublingual salivary gland tissue based on location of the defect. As per the literature overall incidence of Stafne bone defect found range from 0.009% to 0.48%[6].They are more commonly located between premolar and angle region of mandible. Rarely seen in canine and incisor region, where, lesion predominantly found to have male predilection, 8% to 9% cases seen in males in their fifth to seventh decade of life[7]. In our case report it was found on left side of mandible in relation to lateral incisor, canine and premolar.

These defects are believed to be a developmental defect and it do not appear to be present from birth [8]. There are many theories which explain the aetiology of SBDs but there is no universally accepted one. Several studies have suggested that they may be formed by a congenital inclusion of the salivary gland tissues during the development of the mandible. The submandibular gland and parotid gland is related to the posterior variant, sublingual gland and parotid gland is related to the anterior and variant of ascending ramus of the mandible respectively. The other hypothesis are, formation of bone defects as result of pressure atrophy of the submandibular gland, micro trauma caused by the pulsation of the facial artery[1,2, 5]

Nearly 80-90% of cases SBDs are present in the posterior mandible. Anterior lingual salivary gland defects are very rare and presents as non-definitive bony radiolucencies, which makes it more difficult to diagnose [8]. As per the literature the reported incidence of anterior SBDs was found to be range from 0.009% to 0.48% [9]. Stafne's bone cavity in the anterior region of the mandible is first described by Richard and Ziskind in 1957. As per the literature around 33 cases of anterior SBDs are reported so far

and most often they found between canine and premolars. The size of the defect ranges from 0.5 to 2cm [4]. The differential diagnosis for anterior mandible bony defects are radicular cyst, residual cyst and non-inflammatory odontogenic cyst [10].

SBDs are asymptomatic and found accidentally. Similarly in our case, it was found on routine radiographic examination of mandibular fracture. Surgical exploration and biopsy to diagnose the lesion is mandatory. Periodic clinical and radiographic

evaluation is necessary to see the changes in nature and size of the lesion.

Acknowledgment

We would like to thank Dr. Niranjana Kumar, Director; SDM Craniofacial Unit and Dr. Srinath Thakur, Principal; SDM College of Dental Sciences and Hospital for their support, encouragement and facilities provided.

References

1. Sekerci AE, Sisman Y, Etoz M, Aksu Y. European Journal of Radiology Extra Aberrant location of salivary gland inclusion?: Report of a case with review of the literature. *Eur J Radiol Extra* 2011;79:27-31
2. Prechtel C, Stockmann P, Neukam FW, Schlegel KA. Enlargement of a Stafne cyst as an indication for surgical treatment: A case report. *J Cranio-Maxillofacial Surg* 2013;41(3):270-3.
3. Schneider T, Filo K, Locher MC, Gander T, Metzler P, Grätz KW, et al. Stafne bone cavities?: systematic algorithm for diagnosis derived from retrospective data over a 5-year period. *Br J Oral Maxillofac Surg* 2014;52(4):369-74.
4. Courten A, Kuffer R, Samson J, et al. Anterior lingual mandibular salivary gland defect (Stafne defect) presenting as a residual cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:460-64
5. Bornstein MM, Wiest R, Balsiger R, Reichart PA. Anterior Stafne's Bone Cavity Mimicking a Periapical Lesion of Endodontic Origin: Report of Two Cases. *J Endod* 2009;35(11):1598-602.
6. Neville BW, Damm DD, Allen CM, Bouquot JE. *Oral and Maxillofacial Pathology*. 4th ed. Philadelphia: Saunders; 2009
7. Sisman Y, Etoz OA, Mavili E, Sahman H, Tarim Ertas E. Anterior Stafne bone defect mimicking a residual cyst: a case report. *Dentomaxillofac Radiol* 2010;39:124-6.
8. Barak S, Katz J. Anterior lingual mandibular salivary gland defect-a dilemma in diagnosis. *Br J Oral Maxillofac Surg* 1993;31:318-320
9. Philipsen HP, Takata T, Reichart PA, et al. Lingual and buccal mandibular bone depressions: a review based on 583 cases from a world-wide literature survey, including 69 new cases from Japan. *Dentomaxillofac Radiol* 2002;31:281-90.
10. Katz J, Chaushu G, Rotstein I. Stafne's bone cavity in the anterior mandible: a possible diagnostic challenge. *J Endod* 2001;27:304-7.