Variants of major salivary gland related bone defects- A case series

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Abstract

Salivary gland related bone defects or Stafne bone defects are asymptomatic bone cavities in the mandible caused by impingement by the salivary glandular tissue. All the three major salivary glands are in approximation with the mandible and depressions with each can produce radiolucent defects on the surface of the mandible. These bone defects or depressions are categorized as anterior lingual variant, posterior lingual variant and medial ramus variant. These are well defined radiolucent areas that are incidentally discovered on panoramic radiographs and diagnosis may not require elaborative imaging due to asymptomatic nature and characteristic locations. These defects are commonly unilocular but rare multilocular variants have also been described. This case series describe medial ramus variant and 2 posterior lingual variants with one showing multilocular appearance.

Keywords: Bone cysts; Mandibular bone depression; Stafne bone cyst; Salivary gland depression.

Introduction

Salivary gland related defects are the actual defects or the cavities in the mandible that occur due to the cortex bowing inward into the medullary space of the mandible. The cortex is intact separating the medullary space of the mandible from the soft tissues in the glandular space. Since, they are lined by cortex they appear as well defined radiolucent areas with smooth radiopaque rim[1].

The most common major salivary gland related bone defect is an invagination in the medial surface of mandible due to impingement by the lobe of submandibular salivary gland. It was first described by Stafne in 1942[2] and termed as "bone cavities situated near the angle of mandible". The term latent cyst for these was described by Rushton as they appeared cystic on radiographs and were inactive[3]. These have fairly constant location between the inferior alveolar canal and inferior cortical border of mandible at or near the groove made by the facial artery where it crosses the mandible. They are usually unilateral and may show varying shapes with smooth corticated or punched out periphery. They are usually unilocular but rarely may be multilocular.

The anterior variant was first reported by Richard and Ziskind in 1957[4] and are much less common than the posterior variant. They develop to accommodate the smallest major salivary gland i.e. sublingual gland which lies in close proximity to the lingual cortex of the mandible in the canine region. They are seen as well defined radiolucent areas usually 1-1.2cm in diameter superimposing over the root apices of the canine, incisors and rarely premolars mimicking a periapical pathology but the intact lamina dura around the teeth, vitality testing and asymptomatic nature of the lesion makes the diagnosis easier. The lesions may show corticated or punched out margins.

The medial ramus variant was first recognized by Wolf in 1985[4] as depressions on the medial surface of ramus due to depression by the deep lobe of the parotid gland. These are extremely rare with only 17 cases described in literature(6 clinical and 11 archaeological)[5]. They are seen at level with or above the mandibular foramen and may extend superiorly to the base of the condylar neck.

This case series describes 3 cases of major salivary gland related bone defects with multiple variations. The first case demonstrates bilateral posterior lingual variants which is rarely reported. The second case shows a multilocular posterior lingual variant. The third case shows a medial ramus variant in a pediatric patient. All these lesions were incidentally discovered during panoramic radiography of the patient.

Case Description

Case 1: A 60 year old completely edentulous hypertensive male patient reported to the dental OPD for the prosthetic rehabilitation. A scout panoramic radiograph was taken before implant placement which revealed a well-defined unilocular radiolucent lesion in the left angle region below the inferior dental canal roughly triangular in shape measuring approximately 2x1.2 cm with the base of triangle directed at the angle of mandible and the inferior pole partially resorbing the inferior mandibular cortex. The lesion was seen surrounded all around by a radiopaque rim. A very well defined radiolucent area was noted on the posterior aspect of the radiolucency measuring 2mm in diameter suggesting the possible perforation of the buccal cortex also. Interestingly, on the right side, multiple resorptive areas were noted just above the inferior mandibular cortex irregular in shape surrounded by faint corticated borders likely to be bone defects secondary to salivary gland depression. (Fig. 1)

Case 2: A 47 year old male patient reported to the dental opd for the treatment of pain in the right mandibular fixed partial denture in posterior region. A panoramic

radiograph was done which revealed faulty endodontic treatment with periapical abscess irt 46 and mild periodontitis irt 46, 47. An incidental well defined radiolucency was noted on the left side of angle below the crown of horizontally impacted 38(Class III position C). The radiolucency was located below the inferior dental canal and showed a multilocular appearance with size measuring approximately 2 x1.5 cm. It is seen overlapping the superior border of inferior dental canal and partially resorbing the inferior mandibular cortex inferiorly. (Fig. 2)

Case 3: A 9 year old male patient reported to the dental OPD with a history of fall from stairs with consequent pain during mouth opening. A panoramic radiograph was advised to rule out any fracture. The OPG revealed mixed dentition stage with no evident fracture line. On close examination, a very well defined oval shaped unilocular radiolucency was seen in the left ramus above the mandibular foramen measuring about 1.2 x1 cm about 0.5cm from the posterior border of ramus surrounded by a well-defined corticated border.(Fig. 3)

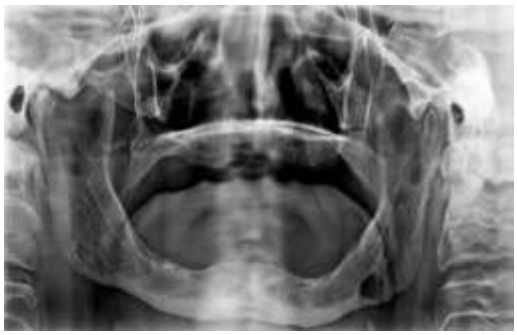

Fig. 1: OPG showing the posterior lingual variant of stafne bone defect on left side

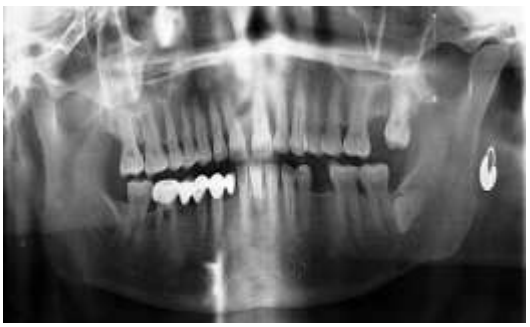

Fig. 2: OPG showing the multilocular posterior lingual variant on left side

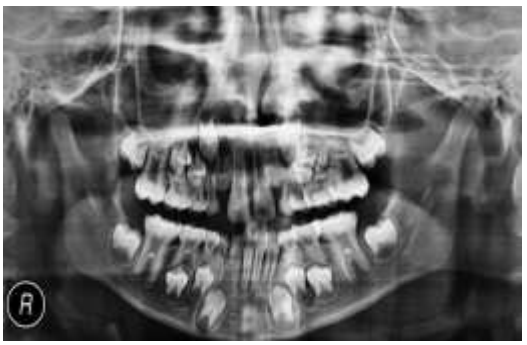

Fig. 3: OPG showing the medial ramus variant on the left side in a pediatric patient

Fig. 4: Diagram showing the various shapes of posterior lingual variants. a) Round b) Oval c) triangular d) Heart shaped

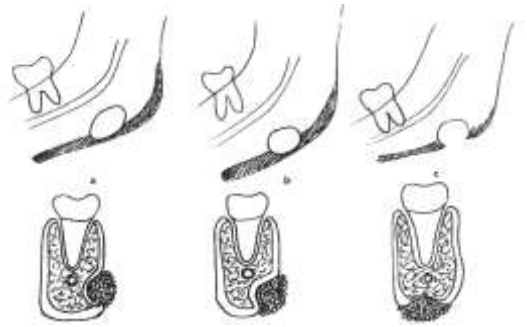

Fig. 5: Diagram showing the lateral and cross-sectional views of the relationship of Stafne bone defect with the inferior mandibular cortex. a) Contact with the cortex b) Partially resorbing the cortex c) saucerization

Discussion

Mandibular bone depressions due to salivary glands have been described through various terminologies like aberrant or ectopic salivary gland defect, static bone defect or cavity, idiopathic bone cavity, latent bone cyst, lingual mandibular bone depression and developmental bone defect[6-9]. The term Stafne bone cyst usually refers to the posterior lingual variant. Instead of aberrant or ectopic salivary gland tissue, they are usually caused by the impingement of bone by the normal salivary gland tissue present in that region. They are usually detected in 5th and 6th decades of life with an incidence of 0.3% in adults[1]. It shows male predilection with a male/female

ratio of 6:1. In our series, all three patients were males and two were in the common age range. However, the medial ramus variant was seen in a pediatric patient which is extremely rare. These lesions are not present since birth and may show some degree of growth to a maximum size of 1-3cm hence, they are called as developmental defects that are relatively static or latent.[10,11] These lesions are usually unilateral but may be bilateral. They are usually unilocular with variable shapes defined in the literature like round, ovoid, triangular and heart shaped.(Fig. 4) The multilocular appearance as seen in our case 2 is extremely rare.

These lesions are usually described on panoramic radiographs and since panoramic radiography involves a negative 8^0 angulation[12], the lingual objects are projected upwards. Hence, these lesions may be seen either contacting or overlapping the inferior cortical border of mandibular canal and very rarely may be seen overlapping both the inferior as well as superior cortical border of mandibular canal. In the first case, it was seen contacting the inferior cortical border of canal and in second case it showed overlapping over both the cortical borders of the mandibular canal.

There can be multiple variations of this posterior lingual variant with respect to the inferior cortical border of mandible. It can be above the cortex, in contact with the superior margin of the cortex, may partially resorb the inferior cortex or may cause saucerization of the entire thickness of cortex.(Fig. 5) The lesion in both the cases showed partial resorption of the inferior cortex with resultant thinning of the inferior cortex.

Similar to this, medial ramus variant may show multiple relations with the posterior surface of ramus with rare lesions producing saucerisation. These lesions are usually round when small and are oval when larger. The margins may be smooth, thick sclerotic or punched out. Our case showed faint corticated periphery on the anterior aspect and punched out on the posterior aspect.

Langlais[4] in their archaeological study of dried mandibles found that smaller lesions show shallow areas of cortical erosion whereas larger lesions show deeper penetrations onto the medullary spaces with better defined margins. Considering this and the radiographic presentation we can state that case 1 showed deeper penetration on the left side as no trabecular pattern was evident in the lesion. It infact showed perforation of the buccal cortex as well, a feature not reported till now. Contrast to this, case 2 and 3 showed internal trabeculations suggesting the shallow nature of these defects.

The most accepted etiology for their embryogenesis is the pressure exerted by the glandular tissue on the medial surface of mandible. However, some investigators have found empty cavities without any glandular tissue during surgical exploration of the area but this can be explained by the accidental displacement of the gland during surgical manipulation. Other authors have also postulated that empty cavities could be due to the defects in the intramembranous ossification of the mandible. Since the location is near the crossing of facial artery over the mandible, it is also postulated that abnormal vascular pressure associated with facial artery could cause necrosis and resorption of the adjacent bone.[2,13,14,15,16] Infact, the influence of arterial pulses can cause bone resorption as patients with hypertension tend to show these bone defects[8,17]. The patient in case 1 was hypertensive and this could explain the well defined defect on left side and probably initiation of a similar defect on the right side that appeared presently as irregular resorptive areas.

Generally, these are composed of salivary glandular tissue but may contain fat, muscle, connective tissue, lymphoid tissue, blood vessels or may have no content at all.

The characteristic location and asymptomatic nature of these lesions usually do not pose any challenges in the diagnosis. Hence, the list of differential diagnosis is limited. The posterior lingual variant is actually the exaggerated version of submandibular gland fossa which is a normal anatomic landmark seen in the posterior mandibular intraoral and extraoral radiographs. It appears as a diffuse radiolucent area below the inferior alveolar canal as compared to the well circumscribed cortical defect in stafne cyst. If the bone defect in posterior variant opens caudally in the inferior mandibular cortex, it appears as saucerization and may mimick subperiosteal lesions causing cupping resorption like subperiosteal neurofibroma. The medial ramus variant may look similar to the medial sigmoid depression. This was first observed on panoramic radiographs by Steven Bricker and was described by Langlais as an anatomic depression on the medial side of the upper ramus just below and anterior to the greatest depth of the sigmoid notch of ramus. They appear as well defined radiolucent areas usually bilateral, less than 5mm in diameter lacking the cortical margin but in an anterior location as compared to medial ramus defect.Since, the anterior lingual variant is superimposed on the dentate region, the various differential diagnosis include periapical abscess, radicular cyst, residual cyst, cement-osseous dysplasias etc. and these are actually difficult to diagnose without the three dimensional imaging.

Clinical diagnosis can also be confirmed by walking a curved needle of 16 gauge along the medial surface of the mandible through either intraoral or extraoral approach[1]. Sialography may be done to show the distribution of the contrast media into the bony defect.Three dimensional imaging in the form of computed tomography and magnetic resonance imaging can be done to show the cortical defect and to evaluate the content of the defect. Since, all the patients in this series were asymptomatic and diagnosis was established based on their characteristic location, CT was not advised due to financial constraints as well as subjecting

the patient for increased radiation exposure was not justifiable.

Hence, this case series aimed to describe various patterns of major salivary gland depressions, the most common of which is posterior lingual variant which in one case showed bicortical involvement a feature not reported till now and other posterior variant showing a rare multilocular variant. The most uncommon variant that is the medial ramus variant is also described for first time in a pediatric patient.

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