Stafne Bone Cavity: A Rare Case Affecting the Anterior Mandible

Annie Pellatt*
Department of Oral and Maxillofacial Surgery, University Hospitals Bristol NHS Trust, UK

Abstract
Stafne’s Bone Cavity (SBC) is a well-documented lesion with clear radiographic diagnostic parameters. Usually occurring in the posterior mandible, the lesion contains salivary gland tissue and is asymptomatic and non-invasive. In contrast, anterior salivary gland inclusion defects are very rare and can present a diagnostic conundrum. They can be unilateral or bilateral, and may appear anywhere between the premolar teeth. The anterior variant is commonly confused with radicular cysts and radiographically can be similar to other insidious lesions. Like their posterior counterparts they occur more often in males in the fifth to seventh decades.

Introduction
This report provides a rare example of a bi-lobed anterior salivary gland inclusion cavity in a 21 year old male. We have provided diagnostic recommendations based on this case and others reported in recent literature.

Case Presentation
A 21 year old male was referred to the maxillofacial team at University Hospital, Bristol following an incidental finding of a bilateral unilocular lesion in the anterior mandible at a dental check. The lesion, which extended from the lower right premolars to the left premolars, was identified on a panoramic film (Figure 1). Apical pathology of odontogenic origin was ruled out with periapical radiographs and pulp sensibility testing. Indeed, all lower anterior teeth were vital. The lesion was asymptomatic. There was no other relevant medical history. A cone beam CT of the mandible was taken demonstrating a partially multilocular defect extending from the lower right five region, crossing the midline into the lower left five regions and enveloping the apices of most of the anterior teeth (Figure 2). The buccal plate was in most part intact, but there was lingual expansion and thinning of the lingual plate (Figure 2). The lesion had a radiographic appearance of giant cell granuloma, keratocyst, ameloblastoma or sialodontogenic tumour. A biopsy of the lesion was performed under general anesthesia. A mucoperiosteal flap was raised lingually where it was evident that there had been perforation of the lingual plate by unidentified soft tissue mass. A biopsy was taken and histopathological analysis confirmed mucous type salivary gland tissue favoring that of the sublingual gland. In conjunction with CT and biopsy an MRI floor of mouth was performed showing a well-defined bilobed soft tissue mass lying anteriorly in the floor of the mouth. The MRI concluded that the lesion represented an anterior variant of a Stafne cyst. The patient was followed up and an MRI repeated for comparison. The lesion had not changed in size and the patient remained asymptomatic.

Discussion
Stafne Bone Cavities (SBCs) are pseudocysts of the mandible. First defined by Stafne in 1942, they are depressions of the lingual cortical bone of the mandible, containing ectopic salivary gland tissue [1]. They appear radiographically as uniloculated, well defined radiolucencies below the inferior alveolar nerve canal in the third molar region [1]. SBCs have an incidence of 0.1% to 0.48%, most commonly occurring in men in the fifth and sixth decades [2,3]. Similar lesions have been identified on the ascending ramus and also lingually in the anterior mandible, although the incidence of these variants is less [4,5]. SBCs are asymptomatic and are usually noted as serendipitous findings during routine dental checks. Stafne defects are generally considered to be a congenital anomaly. It is thought that during mandibular development, part of the salivary gland tissue becomes trapped in the developing mandible. This is consistent with the clinical sign of perforation in the lingual cortex with continuity of the lesion to the adjacent salivary gland. An
alternative theory suggests that over time pressure from adjacent soft tissue structures—such as salivary glands, facial artery or lymphatic infiltration—may lead to development of these bony defects [6,7]. The anterior variant of the Stafne bone cavity can be more accurately referred to as a lingual mandibular salivary gland defect, inclusion or depression [8]. First reported by Richard and Ziskind in 1957, there have been infrequent reports of this anomaly since [9]. A review of the current literature would suggest there have been fewer than 60 cases reported since this date [6,7]. The majority of reports of anterior SBC have described lesions between the canine and lower first molar [10]. The presented case is unusual in that the lesion is bilateral and superimposed over the anterior teeth up the second premolars on either side. A similar case was reported by Kim et al. in 2014 of a bilateral anterior mandibular SBC [6]. Deyhimi et al. [11] 2016, reported an SBC in a similar location, although the lesion was unilateral. Like the posterior variant, anterior SBCs usually occur in males in the fifth or sixth decade. Interestingly we present a case in a 21-year-old man—the youngest reported case to date. The anterior variant of SBC can prove to be diagnostically problematic. Unlike its posterior equivalent, the anterior SBC does not have established radiographic defining characteristics. For example, the posterior SBC is almost always at the angle of the mandible below the IAN, and radiographically can be seen as unilocular. These clear parameters mean SBCs can be confidently diagnosed from clinical and radiographic examination alone. In contrast, anterior SBCs can be unilocular or multilocular, occur anywhere in anterior mandible and can be superimposed over the roots of anterior teeth. They are more often unilateral, but occasionally present as a bilateral lesion. Most commonly they are misdiagnosed as radicular cysts and it is therefore important to exclude dental pathology by carrying out pulp viability tests, such that no unnecessary endodontic treatment is carried out. The anterior variants can also bare resemblance to a number of other pathologies radiographically. Differential diagnoses include fibro-osseous lesions, ameloblastoma, myxoma, central giant cell lesions, odontogenic keratocysts and salivary gland tumors. Therefore, it is recommended that a soft tissue biopsy is taken for an accurate diagnosis of the anterior SBC [11,12].

Conclusion

We have presented a rare case of a bilobed anterior lesion; even more unusual that occurs in a young man of 21. In reporting this case we hope to highlight the key diagnostic principles and features of this rare lesion.

References