Case Report
Idiopathic Benign Hyperplasia of Sublingual Gland: Report of a Case and Review of Literature
Kamath VV, Nagaraja A, Krishnanand Setlur

Abstract
Enlargements of salivary glands have many causes chief among them being, inflammatory, neoplastic or physiological alterations. Non-specific enlargements without any detectable cause have been classified as idiopathic and have also been rarely reported. We describe a case of a 59 year old male patient with a sublingual swelling that was treated with conservative surgical excision. The histopathological diagnosis was that of Idiopathic Hyperplasia of Salivary Gland. The awareness of this lesion is important to highlight the variations in salivary gland pathology and for avoiding diagnosis and treatment pitfalls.

Keywords: Sublingual Gland; Idiopathic; Hyperplasia; Adenomatoid; Salivary Gland; Oncocytes.

Introduction
Pathologies associated with major and minor salivary glands are commonly reported in literature. The most common are infections, inflammations, calculi, cysts and tumors. Physiological enlargements of the salivary glands are secondary to hormonal changes or age-related as in oncocytic hyperplasia. Non-specific enlargements sans physiological or pathological stimuli are rare and hitherto unreported. These enlargements of unknown causes are labeled as Idiopathic Hyperplasia of Salivary Gland. There are no studies in literature listing the prevalence of such occurrences and only isolated case reports are available. We report a case of a 59 year old male with a benign hyperplasia of the salivary gland in the absence of any cause or detectable pathology.

Case Report
A 59 year old male reported to the clinic with complaint of a sublingual swelling on the right floor of the mouth. The swelling was painless, recently increasing in size and caused problems with speech and tongue movements. The swelling was noticed by the patient for the past 8 months. The patient had no significant medical or drug history. Oral examination revealed a lobulated swelling of approximately 5 x 3 cm, freely mobile, non-ulcerated, sessile and arising from the right sublingual gland. No localized lymphadenopathy was observed. There were no history of habits and the right side of the mandible presented with decayed root stumps of the molar teeth (Fig 1). Intraoral and panoramic radiographs revealed no obvious involvement of the mandible. There was no evidence of calcifications of the salivary gland.

The lesion was excised in toto under local anesthesia taking care to avoid damage to the submandibular duct. The patency of the latter was confirmed post excision by cannulation. The excised tissue had the same feel and texture as a normal sublingual gland (Fig 2).

Histopathological examination of the excised mass revealed normal salivary gland tissue with diffuse hyperplasia (Fig 3). Though oncocyes were present there was no oncocytosis. The acini were uniformly enlarged and more in number maintaining the overall architecture of the glandular tissue. Inflammatory infiltration was minimal (Fig 4).

Histopathological examination of the excised mass revealed normal salivary gland tissue with diffuse hyperplasia (Fig 3). Though oncocyes were present there was no oncocytosis. The acini were uniformly enlarged and more in number maintaining the overall architecture of the glandular tissue. Inflammatory infiltration was minimal (Fig 4).

There was also no evidence of calculi or any other pathological strictures of the ducts. No dysplastic changes or abnormalities were noted in the salivary tissue. A histopathological diagnosis of benign hyperplasia of the sublingual gland was returned. The patient has been under observation for two years and there are no signs of recurrence.

Discussion
Enlargements of major and minor salivary glands are common surgico-pathologic conditions encountered by the oral surgeon and pathologist. The common causes of
enlargements include infections, inflammations, cysts, tumors, effects of drugs and metabolic disorders. Idiopathic enlargements of the salivary glands though clinically frequent are reported infrequently in literature. Idiopathic hyperplasia of the sublingual gland (IHSG) is an interesting and rather uncommon presentation seen as a painless, non-neoplastic, non-inflammatory swelling of the sublingual gland clinically mimicking a tumor or a cyst. A literature review indicated only 4 case reports involving a total of 27 cases, excluding the one presented here. The diagnosis of IHSG was defined by one author as the presence of a diffuse, unilateral or bilateral swelling, soft and resilient to palpation at the floor of the mouth. It is imperative that radiologic examination to rule to presence of sialoliths be carried out as routine. A prominent feature of presentation is the partial or total lack of teeth in the region. In their analysis of 25 patients presenting with IHSG, Domaneschi et al found this feature to be statistically significant. The case presented in this report also had a partially edentulous jaw in the region of the swelling with only decayed root stumps of the molar teeth without any sign of infection. Though wearing prosthesis was associated with a few cases of IHSG this did not seem to statistically impose on the etiology of the condition. An analysis of other associated factors like medication, diabetes mellitus and hypertension, infections and HIV did not reveal any correlation with the pathogenesis of the condition.  

Figure 1: Swelling in the right sublingual region, with decayed root remnants of the posterior molar teeth.  

Figure 2: Excised specimen showing normal lobular pattern of salivary gland tissue.  

Figure 3: Photomicrograph of excised tissue showing normal sublingual gland architecture and histology, with hyperplasia. (Hematoxylin and Eosin, X10)  

Figure 4: Note the dilated venules and ducts, lack of inflammation and few oncocyes (Hematoxylin and Eosin, X 10).  

Histologically the tissue is seen to be composed of almost normal appearing mucous acinic with ducts albeit hyperplastic in nature. Mild inflammatory inflammation
and chronic sialadenitis were also reported associated with the hyperplastic tissue. It is of interest to note that a parallel condition of hyperplasia called adenomatous hyperplasia (AH) exists affecting salivary glands but the differentiation is only too obvious. While AH commonly afflicts minor salivary glands especially of the palate, IHSG is a mono-gland affliction. AH afflicts minor mucous salivary glands with the histology indicating hypertrophy and hyperplasia of the minor mucus glands. IHSG is a pan-enlargement of all components of the sublingual gland especially the acini and ducts. Tagawa et al described an enlargement of the sublingual gland in a patient who was being treated with calcium channel blockers. Pathological examination of the gland tissue revealed multiple normal appearing serous and mucous acini but with the ratio of serous to mucous acini being 7:1. There have been no further confirmations of such an exclusive serous acinar hyperplasia in the literature. The authors called this lesion “Adenomatoid serous lobular hyperplasia of the sublingual gland” and attributed the enlargement to the probable use of calcium channel blockers.

AH has been known to be confused with a minor salivary gland tumor; interestingly in one FNAC study the authors state that there were indications of the lesion mimicking a mucoepidermoid carcinoma.

AH has been strongly associated with local chronic trauma caused by dentures and the habit of smoking while no such known factors are associated with IHSG except the observation of partial or total edentulousness. Table 1 lists the major differences between the two conditions. The establishment of IHSG thus needs to be recorded and the condition recognized to avoid diagnostic pitfalls. It was interesting to note in the study of the 25 cases by Domaneschi et al, only six cases were punch biopsied and the authors’ emphasis on avoidance of surgical treatment for the condition. The presentation of the IHSG condition as a swelling mimics a tumor or a cyst and the patient is most often apprehensive if not overtly cancerophobic.

A strong case can be made for surgical excision of the lesion for various reasons. Firstly total excision renders the whole tissue available for histological evaluation thus ruling out pockets of tumor transformation if any. Secondly the procedure is relatively minor, feasible in the normal surgical clinic environment and with minimal morbidity. Thirdly no long term studies exist regarding any change in such initially diagnosed IHSG lesions if left untreated. It would thus be prudent to carry out a surgical excision of the lesion.

The pathogenesis of the condition is still unknown. Though many associated factors like medication for systemic illnesses, prosthesis usage and habits have been recorded none seems to fit the bill as a similar group of individuals associated with the factors did not show the development of IHSG. The lack of teeth in the region seems to be a common persistent factor in almost all case reports. It may be conjectured that the additional space created by the edentulousness and the movements of the tongue in an extended area tend to push the sublingual tissues along with the gland resulting in a chronic low grade irritation resulting in hyperplasia; almost similar to masseteric hypertrophy seen in individuals with clenching and bruxism. Interestingly Filho et al reported a similar condition of idiopathic enlargements of the sublingual gland involving 24 cases. They labeled this condition “mouth floor enlargements (MFE)”. The histological presentation of their cases varied considerably. Six of the 24 sublingual glands removed were microscopically normal, while the other specimens presented acinar atrophy with hyperplasia of duct-like structures. Interstitial fibrosis was observed in 18 cases and was accompanied by adipose tissue infiltration in 15. Decreased lymphoid tissue was observed in 16 samples and oncocytosis was present in 5 cases. The major differences between the MFE cases and IHSG seem to be the histology.

The authors debate on an appropriate nomenclature for this entity and rule out the use of the term “hyperplasia” due to the conflicting observation of atrophy in some cases. They also postulate that the altered attachment of the mylohyoid muscle and the partial or total edentulousness may have contributed to the non-specific enlargement of the sublingual glands.

While no cases have been reported undergoing tumorigenic transformation it would be prudent to recognize the condition as a potential hazard and initiate surgical intervention; more so to enable a clear area for future prosthetic rehabilitation and removal of the mass.
### Table 1: Comparison between Adenomatous Serous Hyperplasia and Idiopathic Hyperplasia of Salivary Gland

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Adenomatous Serous Hyperplasia (ASH)</th>
<th>Idiopathic Hyperplasia of Salivary Gland (IHSG)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Histology</strong></td>
<td>1. Ductal Hyperplasia and Hypertrophy</td>
<td>1. Generalized hyperplasia of sublingual gland elements</td>
</tr>
<tr>
<td></td>
<td>2. May mimick a low grade mucocoeidermoid carcinoma histologically and in FNACs</td>
<td>2. No specific relation to any salivary gland tumor.</td>
</tr>
<tr>
<td><strong>Associated Cause</strong></td>
<td>Smoking, trauma, use of calcium channel blockers</td>
<td>None except for partial or total edentulousness.</td>
</tr>
<tr>
<td><strong>Site affliction</strong></td>
<td>Usually afflicts minor salivary glands of palate</td>
<td>Mono-gland affliction, usually sublingual</td>
</tr>
</tbody>
</table>

Table 1: Comparison between Adenomatous Serous Hyperplasia and Idiopathic Hyperplasia of Salivary Gland.

**Author Affiliations**

**Acknowledgement**
We would like thank all the staff members of the Department of Oral and Maxillofacial Pathology for their support.

**References**

**Corresponding Author**
Dr. Kamath VV, MDS, FDSRCS, FFDRCSI, Professor and Head, Department of Oral and Maxillofacial Pathology, Dr Syamala Reddy Dental College, Munnekolala, Marathalli, Bangalore-560037, India. E-mail: kamathvv2010@gmail.com Mobile: 09845037021

Source of Support: Nil, Conflict of Interest: None Declared.