Intraductal Papilloma of the Buccal Mucosa Salivary Gland

Shin-ichi Yamada,1 Souichi Yanamoto,2 Goro Kawasaki,1 Akio Mizuno,1 Shuichi Fujita,2
Tohru Ikeda2
1Department of Oral and Maxillofacial Surgery, Unit of Translational Medicine, and 2Department of
Oral Pathology and Bone Metabolism, Unit of Basic Medical Sciences, Nagasaki University
Graduate School of Biomedical Sciences, Nagasaki, Japan

Abstract

Salivary gland papillomas are rare tumours arising from the ductal epithelium. Intraductal papillomas,
the most rare of all ductal papillomas, have been reported to occur almost exclusively in the excretory
ducts of the minor salivary glands. An intraductal papilloma of the left buccal mucosa in a 51-year-old
man is reported. Histology of resected tumour revealed that the duct had dilated to form a thick-walled
cyst that contained the papillary tumour with intracystic endoluminal proliferation. No signs
of recurrence were evident 8 months after surgery.

Key words: Mouth mucosa, Papilloma, intraductal, Salivary gland neoplasms, Salivary glands, minor

Introduction

Salivary gland ductal papillomas are rare benign tumours. They have been categorised into intraductal papilloma,
inverted ductal papilloma, and salivadenoma papilliferum. Intraductal papillomas are the most rare of all ductal
papillomas and were first reported by Castigliano and Gold1 in 1954 as a lesion arising in the hard palate. According to
Seifert et al,2 the typical intraductal papilloma contains only intraductal papillary growths of columnar or squamous
epithelial cells with connective tissue cores. With the exception of a few cases,3-6 this tumour occurs in the minor
salivary glands.1,8,9,10-12 Histologically, intraductal papillomas consist of papillary intraductal projections with connective
tissue cores extending into widely dilated ducts or cystic spaces.2 We describe a rare case of intraductal papilloma
arising in a buccal minor salivary gland.

Case Report

A 51-year-old man came to our clinic with a 2 1/2-year history of a left cheek mass. Past medical history was unremarkable.

Physical examination revealed a diffuse swelling superior to the left lateral angle of mouth. Intraoral examination
demonstrated a 1.8 x 1.5 x 1.2 cm, relatively well-defined, elastic, hard, purplish-blue mass in the left buccal mucosa

(Figure 1). The mucosa over the mass was intact and movable, while the compressibility of the mass was unclear.

Although fine-needle aspiration of the lesion was attempted for cytological diagnosis, nothing could be aspirated. No
palpable regional lymphadenopathy was evident.

Magnetic resonance imaging scan was performed for the differential diagnosis and preoperative evaluation of the mass. T1-weighted images revealed a well-circumscribed solitary mass, while T2-weighted images demonstrated the mass to have a high intensity (Figure 2a and Figure 2b). The mass was 1.5 cm in diameter and partially lobular. Clinically and radiographically, a tentative diagnosis of benign tumour of the buccal mucosa was made.

Correspondence:
Shin-ichi Yamada, DDS, PhD, Department of Oral and Maxillofacial Surgery, Unit of Translational Medicine, Course of Medical and Dental Sciences, Nagasaki University Graduate School of Biomedical Sciences, 1-7-1 Sakamoto, Nagasaki 852 8588, Japan.
Tel: (81 95) 849 7666; Fax: (81 95) 849 7700;
E-mail: shishin@nagasaki-u.ac.jp

Figure 2. Axial magnetic resonance images of the lesion. (a) T1-weighted image revealing a well-circumscribed solitary mass in the left buccal region. (b) T2-weighted image showing the tumour to have high intensity. The mass was 1.5 cm in diameter and partially lobular.

Figure 3. Excised intraductal papilloma. (a) The tumour was excised together with overlying mucosa and surrounding normal tissue. (b) The cut surface of the excised specimen revealed a unilocular cystic mass containing a small amount of dark brown muddy fluid.

Excision was performed under general anaesthesia. During the operation, the tumour was resected together with the overlying mucosa and surrounding normal tissue (Figure 3a). The cut surface of the excised specimen revealed a unilocular cystic mass containing a small amount of dark brown muddy fluid (Figure 3b). Histological examination revealed that the duct had dilated to form a thick-walled cyst containing papillary projections with intracystic endoluminal proliferation (Figure 4). The neoplastic papillary projections had central fibrovascular cores and were covered by a single or double layer of columnar or cuboidal epithelium (Figure 5). Some blood vessels in the fibrous cores lined by tumour cells were dilated and filled with erythrocytes. Thick fibrous cores had undergone hyaline changes with small spherical calcifications (Figure 6). Mucicarmine staining demonstrated scattered mucous cells intermingled in the tumour (Figure 7). No nuclear atypia or mitoses were seen. On the basis of these findings, the lesion was histopathologically diagnosed

Figure 4. Intraductal papilloma within the dilated duct. Complicated branching papillary proliferative projections were observed in the dilated duct, giving the appearance of a unilocular cyst (haematoxylin and eosin, × 40).
as an intraductal papilloma. No signs of recurrence have been observed 8 months after surgery.

Discussion

Intraductal papillomas of the salivary glands are rare benign tumours. According to a review conducted by the Armed Forces Institute of Pathology, only 4 intraductal papillomas were identified among nearly 3100 cases of epithelial salivary gland tumours. The reported intraductal papillomas have mainly been found in the minor salivary glands — in the palate, buccal mucosa, lip, and tongue. However, they have also been reported in the parotid gland, sublingual gland, and submandibular gland ducts (Table 1).

Histologically, intraductal papillomas characteristically exhibit ductal dilatation and papillary projections into the lumen, forming a unicystic structure. Microscopic analysis has found these tumours to originate within the main excretory ducts. In an immunohistochemical study, Ishikawa et al. reported that intraductal papillomas had epithelial origins from the excretory salivary gland ducts, and that the tumour cells exhibited secretory potential. In addition, ultrastructurally, these tumour cells of epithelial origin exhibit many microvilli on their luminal surface and rough endoplasmic reticulum, numerous Golgi apparatus, and mitochondria in the cytoplasm. These findings also suggest that the epithelial cells of intraductal papillomas have high secretory potential.

Clinically, intraductal papillomas have been preoperatively misdiagnosed as other tumours. Fine-needle aspiration has, therefore, been performed for preoperative diagnosis; however, because intraductal papillomas are usually small cystic formations of the salivary duct, the aspiration of typical papillary cells is challenging and it is often difficult to make a preoperative diagnosis. We encountered this problem with the present case; although we performed fine-needle aspiration, no fluid could be aspirated.

The differential diagnosis of intraductal papilloma includes sialadenoma papilliferum, papillary cystadenoma, and inverted ductal papilloma. Sialadenoma papilliferum is an exophytic mucosal lesion of minor salivary glands in which papillary projections of stratified squamous epithelium form in tortuous dilated excretory ducts. Papillary cystadenoma has been described as a cystic adenoma in which multiple cystic spaces are filled with papillary projections. Inverted ductal papillomas arise from the superficial portion of the salivary gland excretory duct, with the squamous epithelial cells extending into the surrounding connective tissue.
### Table 1. A review of the literature of intraductal papillomas.

<table>
<thead>
<tr>
<th>Reference</th>
<th>Year</th>
<th>Age/gender</th>
<th>Location</th>
<th>Size (cm)</th>
<th>Clinical appearance</th>
<th>Treatment</th>
<th>Prognosis (follow-up period)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Castigliano and Gold⁵</td>
<td>1954</td>
<td>58/male</td>
<td>Palate</td>
<td>0.8</td>
<td>Ulcer</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td>Abbey⁵¹</td>
<td>1975</td>
<td>50/female</td>
<td>Upper lip</td>
<td>1.0 × 1.0</td>
<td>Round, firm, elevated mass</td>
<td>Excision</td>
<td>NED (6 months)</td>
</tr>
<tr>
<td>King and Hill⁶</td>
<td>1993</td>
<td>87/male</td>
<td>Parotid gland</td>
<td>2.0 × 2.0</td>
<td>Well-circumscribed firm mass</td>
<td>Partial parotidectomy</td>
<td>NED (6 months)</td>
</tr>
<tr>
<td>Ishikawa et al⁶⁰</td>
<td>1993</td>
<td>58/female</td>
<td>Blandin-Nuhn’s gland</td>
<td>1.5 × 0.7</td>
<td>Asymptomatic smooth mass</td>
<td>Excision</td>
<td>NED (2 years)</td>
</tr>
<tr>
<td>Alho et al⁴</td>
<td>1996</td>
<td>50/female</td>
<td>Parotid gland</td>
<td>2.0 × 2.0</td>
<td>Round tumour</td>
<td>Parotidectomy</td>
<td>NED (3 months)</td>
</tr>
<tr>
<td>Hara et al⁵</td>
<td>1999</td>
<td>72/female</td>
<td>Oral floor</td>
<td>3.0</td>
<td>Lanula-like lesion</td>
<td>Sublingual glandectomy</td>
<td>NS</td>
</tr>
<tr>
<td>Soofer and Tabbara⁴</td>
<td>1999</td>
<td>55/female</td>
<td>Parotid gland</td>
<td>2.0</td>
<td>Firm, non-moveable mass</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td></td>
<td></td>
<td>49/male</td>
<td>Submandibular gland</td>
<td>5.3</td>
<td>Firm mass</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td>Mirza et al⁵</td>
<td>2000</td>
<td>76/male</td>
<td>Submandibular gland</td>
<td>3.0</td>
<td>Swelling</td>
<td>Excision</td>
<td>NED (NS)</td>
</tr>
<tr>
<td>Nagao et al⁶⁰</td>
<td>2000</td>
<td>72/male</td>
<td>Oral floor</td>
<td>2.0</td>
<td>Round mass</td>
<td>Partial sublingual glandectomy</td>
<td>NED (2.5 years)</td>
</tr>
<tr>
<td>Brannon et al⁵</td>
<td>2001</td>
<td>77/male</td>
<td>Buccal mucosa</td>
<td>NS</td>
<td>Painless, well-defined submucosal mass</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td></td>
<td></td>
<td>50/female</td>
<td>Parotid gland</td>
<td>NS</td>
<td>Painless, well-defined submucosal mass</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td></td>
<td></td>
<td>61/male</td>
<td>Upper lip</td>
<td>NS</td>
<td>Painless, well-defined submucosal mass</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td>Iguchi et al⁵</td>
<td>2002</td>
<td>41/female</td>
<td>Parotid gland</td>
<td>1.0</td>
<td>Elastic, hard, round, smooth-surfaced mass</td>
<td>Extirpation</td>
<td>NED (5 months)</td>
</tr>
<tr>
<td>Fattelli et al⁵</td>
<td>2002</td>
<td>74/female</td>
<td>Palate</td>
<td>1.0</td>
<td>Sessile lesion</td>
<td>Excision</td>
<td>NED (3 years)</td>
</tr>
<tr>
<td>Obara et al⁶⁰</td>
<td>2004</td>
<td>65/male</td>
<td>Upper lip</td>
<td>2.2 × 1.5</td>
<td>Smooth, elastic, soft swelling</td>
<td>Excision</td>
<td>NS</td>
</tr>
<tr>
<td>Ripari et al⁶⁰</td>
<td>2005</td>
<td>26/male</td>
<td>Buccal mucosa</td>
<td>2.0</td>
<td>Bilobular, round mass</td>
<td>Excision</td>
<td>NED (NS)</td>
</tr>
<tr>
<td>Present case</td>
<td></td>
<td>51/male</td>
<td>Buccal mucosa</td>
<td>1.8 × 1.5</td>
<td>Well-defined, elastic hard, purplish-blue mass</td>
<td>Excision</td>
<td>NED (8 months)</td>
</tr>
</tbody>
</table>

Abbreviations: NS = not specified; NED = no evidence of disease.

Conservative local excision appears to be adequate and curative for intraductal papilloma. Although the tumour is generally considered benign, the literature contains 2 cases of malignant salivary tumours that appeared to be related to intraductal papillomas.⁶⁰ In the first case, papillary adenocarcinoma probably arose from an intraductal papilloma of the parotid gland, while in the second, a parotid gland tumour with lymph node metastasis was considered to have arisen from an intraductal papilloma as the malignant counterpart.⁶⁰ According to a comparison of the immunohistochemical findings of benign intraductal papilloma and malignant intraductal papilloma, both tumours demonstrate properties of salivary duct luminal cells. However, the malignant tumour had a much higher Ki-67 labelling index (defined as the percentage of Ki-67-positive cells determined by counting at least 1000 tumour cells), and positive reactivity for p53 and carcinoembryonic antigen, indicating greater cell proliferation.⁶⁰

In conclusion, we report a rare case of intraductal papilloma arising in the buccal mucosa that was treated by simple excision.