



Case report

# Intraoral intraductal papilloma: a case report

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## Intraoral intraductal papilloma: a case report

Ductal papillomas have unique papillary features arising from the salivary gland duct system. It comprises three rare benign adenomas, namely, inverted ductal papilloma, sialadenoma papilliferum and intraductal papilloma. Here the first case of intraductal papilloma developed in the minor salivary gland of the vestibule of the oral cavity in a 71-year-old Chinese female living in a nursing home is described. This case is worthy of clinical investigation as it presents as an intraoral swelling and is mistakenly regarded as the result of a periapical pathosis. It also emphasises that a nurse or an oral hygienist who is usually the first-line oral carer of the residents of a nursing home, should be trained to perform the daily dental check and request a dentist's services when necessary.

**Keywords:** intraductal papilloma, oral care, vestibule.

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## Introduction

Intraductal papilloma is a benign papillary lesion originating from the salivary gland duct system with most cases occurring in geriatric patients<sup>1</sup>. However, a few paediatric cases have also been reported<sup>2-4</sup>. Overall, the minor salivary gland is more frequently involved than the major gland<sup>1</sup> and clinically, is usually a painless, well-defined submucosal mass<sup>1</sup>. The first case of a biopsy-proven intraductal papilloma occurring in the vestibule of the oral cavity is reported.

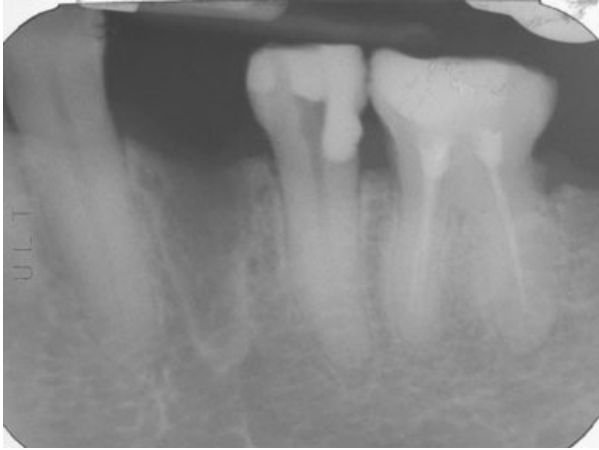
## Case report

A 71-year-old Chinese woman, functionally dependent and living in a nursing home, was referred to our institution from a local practice dentist, complaining of a firm, painless, non-tender submucosal mass for approximately 12 months. The mass was approximately 0.8 cm in diameter, with normal overlying mucosa and was located in the left lower vestibular area, opposite the lower second bicuspid and first molar (Fig. 1). About 1 year previously, a nurse at the patient's nursing home noted this swelling at routine oral health care and the patient was subsequently referred to a local practice dentist who regarded this mass as end-

odontic in origin due to a periapical pathosis of the left lower second bicuspid, as observed from a periapical radiograph (Fig. 2). However, the swelling was still present without obvious alteration in size after the completion of endodontic treatment of this tooth. Consequently, a benign mesenchymal tumour was suspected and the whole tumour was removed under local anaesthesia. The patient denied any oral habits including cigarette smoking.



**Figure 1** A submucosal swelling with normal overlying mucosa in the left lower vestibule, opposite the left lower second bicuspid and first molar.



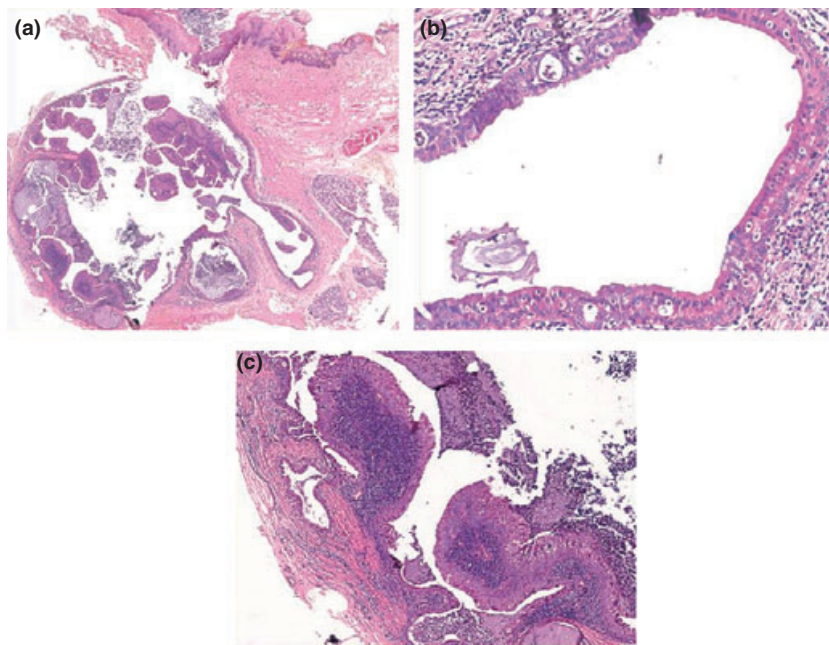
**Figure 2** Periapical film revealing a periapical pathosis over the left lower bicuspid and hypercementosis for the left lower molar.

Microscopic examination revealed a wedge of tissue covered by keratinised stratified squamous epithelium. A cystic space, exhibiting many papillary infoldings of the epithelium, was present within the fibrovascular connective tissue stroma with multiple lobules of mucous acini and fragments of skeletal muscles (Fig. 3a). This cystic space was lined by columnar/cuboidal eosinophilic epi-

thelial cells with goblet cells (Fig. 3b). Subjacent to the lining epithelium was a lymphocytic infiltrate with several well-formed follicles (Fig. 3c). Therefore, a histological diagnosis of an intraductal papilloma with secondary inflammation was rendered.

## Discussion

This case fulfils the histological criteria of being an intraductal papilloma with the exception of the presence of secondary inflammation. The exact incidence of intraductal papilloma is not easy to evaluate in the literature due to the inconsistent terminology as the term 'papillary cystadenoma' is often applied instead of the term 'intraductal papilloma'. Nevertheless, according to a recent review on salivary ductal papillomas by Brannon *et al.*<sup>5</sup>, at least 40 cases have been described in both the minor and major salivary glands. Reviewing the English language literature, intraductal papillomas have been documented in the minor salivary glands of the lip (13 cases), the buccal mucosa (seven cases), the palate (four cases), tongue (one case), and oral cavity with the location not otherwise specified (one case)<sup>1,5-11</sup>. Consequently, the present case report may be the first description of a lesion of the intraductal papilloma in the vestibule of the oral cavity.



**Figure 3** Histological examination. (a) Underlying the surface keratinised squamous epithelium, there is a cystic space with many papillary epithelial infoldings. Multiple lobules of mucous acini, fragments of skeletal muscles are also present within the fibrovascular connective tissue stroma (haematoxylin and eosin stain, H&E, 10×). (b) This cystic space is lined by a row of columnar/cuboidal eosinophilic epithelial cells (H&E, 100×). (c) Subjacent to the lining epithelium is a lymphocytic infiltrate (H&E, 100×).

Besides the aforementioned pathological interest, the current case is also of clinical significance for a swelling, although without an obvious symptom/sign a swelling should be subject to periodic follow-up, especially, as demonstrated in this case, when it still persists after endodontic treatment. Such a patient should be referred to an oral pathologist or oral surgeon for a further clinical check-up and biopsy examination. Indeed, the rare occurrence of a malignant tumour originating from an intraductal papilloma of the major salivary glands has been reported<sup>12,13</sup>. Therefore, a histological examination is very important for an exact diagnosis.

With prolonged life expectancy, elderly individuals with compromised independence may choose to reside in a residential home even whilst a certain level of self-care is possible. However, functionally dependent elderly individuals, like the patient in this report, may need nursing and personal care when admitted to nursing homes where all their care needs are assumed to be met. Then, a nurse or an oral hygienist is usually the first-line oral carer of the residents<sup>14</sup> and he or she must be trained to carry out routine dental check and request a dentist's services for the elderly resident where necessary.

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