CASE REPORT

The use of intraoral ultrasound in the characterization of minor salivary gland malignancy: report of two cases

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It is generally accepted that ultrasound is now the first line of imaging of palpable lumps of the neck. Standardized protocols exist for the evaluation of thyroid, salivary gland and nodal disease, and sonography is increasingly being used in the characterization of intraoral soft tissue lesions. Here, we present two cases where intraoral sonography was invaluable in the early detection of oral malignancy.

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Introduction

Diagnostic imaging with CT, MRI and conventional (transcutaneous) ultrasound of tumours of intraoral minor salivary glands can be challenging because of hard-tissue anatomy, movement and the presence of dental restorations.

However, early diagnosis is imperative, as at least half of these will follow an aggressive (malignant) course.

Here, we report two cases where intraoral ultrasound was used in the pre-biopsy characterization of minor salivary gland malignancy.

Case reports

Case 1

A mildly obese, 60-year-old Caucasian male with an arthralgic gait was referred by his general dental practitioner, with an unusual cystic swelling in the right retromolar trigone. The patient was a non-smoker and an occasional user of alcohol. The swelling was asymptomatic and the patient was unaware of its presence. Medical history included well-controlled hypertension, angina, asthma and type II diabetes, and the patient's mobility was restricted by excess weight and recent right knee and left hip replacements. His medications comprised indoramin,

ramipril, metformin, gliclazide, amlodipine, ferrous sulphate, atenolol, isosorbide mononitrate, tamsulosin, tramadol and glyceryl trinitrate tablets and spray. His dental history was of extensive, mainly amalgam, restorations.

Intraoral examination revealed a soft, bilobed cystic swelling with prominent varicosities distal to the lower right third molar tooth. This was 3 cm in diameter and although superficially fluctuant, it appeared to be fixed to the underlying retromolar pad (**Figure 1a**). There were no palpable lymph nodes, and cranial nerve function was intact. As the margins of the lesion were not identifiable by clinical means alone, imaging investigations were initiated. Unfortunately, the patient was unable to tolerate MRI (claustrophobia and discomfort), and contrast-enhanced CT was justified as an alternative.

Surprisingly, given the clinical presentation, CT findings were reported as inconclusive, with beam-hardening streak artefacts from dental restorations obscuring much of the region of interest (**Figure 1b**).

In view of the limited findings from CT and the suggestion of varicosities posing a biopsy risk, pre-operative power Doppler ultrasound was performed as a means of assessing vascular flow rate. The lesion was too deeply seated for ultrasound penetration from extraoral sites at the edge of the mandible; thus, an intraoral probe (4–8 MHz; Hitachi Medical Systems, Wellingborough, UK,

Figure 1 (a) Photograph of Case 1 swelling *in situ*, and (b) axial contrast-enhanced CT showing the region obscured by beam-hardening artefact.

on a Hitachi 850 platform) was employed using saliva as a coupling medium.

This revealed the mass to be well defined with two distinct compartments. Echogenic content in the superficial compartment was thought to be complex fluid, but the hypoechoic homogeneity in the deep aspect suggested a solid component (Figure 2a). The full extent of the lesion was defined, and minimal colour Doppler flow values provided the reassurance for safe biopsy.

Direct fine-needle aspiration (FNA) cytology performed prior to imaging identified foamy macrophages,

cholesterol crystals and blood components and was deemed inconclusive. At this time, the solid component had not been identified.

Subsequent incisional biopsy revealed the lesion to be a low-grade mucoepidermoid carcinoma (Figure 2b). This histological diagnosis was consistent with the lesional characteristics observed on ultrasound. A fixed lesion of oral mucosa, with substantive solid and fluid compartments, is most probably a salivary neoplasm. Well-defined margins, high proportion of cystic to solid component and low vascularity would further characterize this as a benign

Figure 2 (a) Ultrasound image of intraoral lesion (Case 1) taken using high-frequency intraoral probe. Real-time imaging of the superficial lobe (white arrow), reveals a thick fluid content. White dots demonstrate the cystic components, with white crosses in the positions of the measuring calipers. DI demarcates the deeper, more solid component of the lesion. (b) Photomicrograph of a section from the excised cyst shows a mucinous content (black arrow) and complex epithelial wall (white arrow), including polygonal cells, mucous, goblet and clear cells. Features are consistent with a low-grade mucoepidermoid carcinoma.

lesion or low-grade malignancy. As the mass was small and superficial, laser resection was carried out with minimal post-operative discomfort and no recurrence after 3 months.

In this case, all of the imaging information about the lesion was provided by ultrasound. Not only were the full extent and relations defined for operative guidance, but also the behavioural characteristics in determining the management pathway.

Case 2

A 51-year-old Afro-Caribbean male presented at a dental accident and emergency department. The patient was an occasional smoker and consumer of alcohol, and his chief complaint was of a slowly growing, painless lump in his palate, which had been present for 3 months. He had no significant medical history, and he was not on any medications and a dental history comprising only a few small restorative procedures.

On examination, there was a 2 × 3-cm diffuse, erythematous, exophytic mass at the junction of the hard and soft palates. As an adjunct to the clinical examination, a high-frequency, intraoral “hockey-stick” probe (Philips intraoperative probe 7–15 MHz on a Philips iu22 platform with saliva as a coupling agent) was employed to determine the sonographic characteristics of the mass, prior to biopsy.

The lesion was solid, of heterogeneous echogenicity and involved the bony floor of nose/maxillary antrum. Its intraoral dimensions (1.5 × 1.4 cm) were estimated, as the lesion was not well defined. Furthermore, it was unclear how far the lesion extended into the maxillary antrum or nasal fossa (Figure 3a). Doppler flow demonstrated the lesion to be highly vascular, with many pulsatile vessels (Figure 3b). The size, poorly defined margin, mixed echogenicity and aberrant vasculature were highly suspicious of malignancy; thus, MRI and incisional biopsy were expedited.

The histological features were consistent with those of an adenoid cystic carcinoma (Figure 4). The biopsy report confirmed the ultrasound characterization that

this was an aggressive, infiltrative lesion with abundant vasculature. No discrete mass was identified, but the sample was scattered with islands of lesional tissue, each associated with a new vessel. Fortunately, MRI showed that there was limited (if any) nasal invasion, perineural spread or lymphadenopathy (Figure 5a,b), and the complete surgical resection was affected. Although the long-term follow-up of adenoid cystic carcinoma is essential,¹ 6-month review revealed no recurrence.

Discussion

High-frequency ultrasonography is the modality of choice for imaging palpable soft tissue swellings of the face and neck.² Its near-field spatial resolution is superior to both CT and MR, ideally suiting the neck, as most structures and pathoses lie between 1 and 5 cm below the skin surface. Reducing the probe frequency increases this imaging depth (for example in parotid gland evaluation), but the intact ossified facial skeleton is impenetrable to ultrasound, bringing the examination of the deep face to an abrupt halt when using the conventional transcutaneous route.

With resourceful probe positioning, even this barrier can be somewhat circumvented, by imaging from within the oral cavity. Many otolaryngologists, for example, have extended their clinical examination of the oropharynx, and parapharyngeal and peritonsillar spaces, with small footprint intraoral ultrasound probes.³ Employing local anaesthesia to manage the gag reflex, ultrasound-guided localization, characterization and aspiration in these regions are proving very useful indeed.⁴

More anteriorly, the use of ultrasound in dentistry is also a gathering pace.⁵ Whilst ultrasound as an alternative to ionizing radiation in the diagnosis of caries and periodontal disease may still be in its infancy, sonographic differentiation of dental tissues has been demonstrated in the mouth, using carefully positioned compact (hockey-stick) probes.⁶

Figure 3 Intraoral ultrasound, using high-frequency “hockey-stick” probe, of a swelling in the midline of the palate (Case 2). Before (a), and after (b) colour Doppler was applied (white arrows indicate the surface of the hard palate). X, presumed sonographic depth of the lesion.

Figure 4 Photomicrograph of a section following excision of the swelling (hemimaxillectomy) showing adenoid cystic carcinoma infiltrating as cords and islands (black arrows) within salivary tissue (mucous acini marked by white arrows) (thanks to Dr Gayani Pitiyage).

These custom-designed probes exhibit manoeuvrability comparable to dental handpieces and are used by radiologists to examine the intraoral component of parotid and submandibular ducts. Together with extraoral sonography, these provide an alternative to conventional and MR sialography in identifying salivary calculi.⁷ In those facilities where such a probe is available, its use has been extended to the imaging of suspicious oral mucosal lesions prior to biopsy and MRI or CT. With these transducers, multiplanar lesion dimensions, margins and blood supply can be assessed, cystic lesions can be distinguished from solid ones and calcifications can be identified. FNA cytology, core biopsy and examination of superficial cervical lymph nodes can also be included within the examination in a “one-stop” clinic setting.

Mucosal lesions of the oral tongue (anterior two thirds) and floor of the mouth are readily examined by

intraoral ultrasound.⁸ Indeed, owing to its superior near-field resolution, sonographic evaluation of tumour thickness of squamous cell carcinoma is more accurate than any other imaging measure. This has significance for carcinoma *in situ*, as the risk of microscopic lymph node metastasis increases significantly above 7 mm.⁹ Bone invasion of the jaws can be detected, and sonographic appearances distinguishing invasion or remodelling of the palate have been previously described.¹⁰

Here, we described the diagnostic efficacy of ultrasound for pre-biopsy characterization of two cases of minor salivary gland malignancy. Although minor salivary gland neoplasia is rare, constituting <0.5% of head and neck tumours, 50% of these are likely to be malignant, making early diagnosis imperative.^{11,12}

The diagnosis in Case 1 was of low-grade mucoepidermoid carcinoma. Presenting clinically as a benign looking mucocele of the retromolar fossa, its deeper intraosseous component was only identified by means of intraoral ultrasound. Following the initial inconclusive FNA, an incisional biopsy proved diagnostic. The sonographic characteristics of a well-defined, relatively avascular cystic and solid lesion were in keeping with the grading markers used for histopathological diagnosis.¹³

Conversely, the torrid vascularity and poor definition of the palatal lesion in Case 2 indicated a more aggressive behaviour, which was similarly confirmed by the histological report of the biopsy from this adenoid cystic carcinoma.

The soft tissue contrast (resolution) of MR has hitherto rendered it as the gold standard imaging modality for oral mucosal lesions. Using multiple sequences and planes and with the use of saturation slabs and contrast agents, tissue characteristics are determined by changes in signal intensity with change in sequence. Conventionally, malignancy is a biopsy-proven diagnosis, performed at tissue and cellular levels. The usual role of imaging is in defining the extent of a lesion, evaluating lymphadenopathy and staging.

Figure 5 T_1 gadolinium contrast-enhanced MRI showing heterogeneous contrast enhancement in the site of the swelling (marked with arrows): (a) axial and (b) coronal images.

However, this should be performed prior to biopsy to avoid confusion from any accompanying inflammation.

At the macroscopic level of MR or ultrasound, tissue characteristics additionally inform upon the behaviour of neoplastic lesions. Histopathological criteria such as the zone of transition between normal and abnormal tissue, proportion of extracellular matrix and vascular elements are all demonstrable by imaging and may help to inform the decision of excisional or incisional biopsy.

MR cannot be used in all individuals, and as with CT, the diagnostic yield of intraoral imaging is adversely affected by the presence of dental restorations. For example, MRI could not be carried out in Case 1, and contrast-enhanced CT proved to be an ineffective alternative, as the lesion was not even identified. This was due to beam-hardening effect from amalgam restorations. In hindsight, this might have been marginally reduced by tilting the gantry,¹⁴ but intraoral ultrasound proved to be an effective alternative, with no restorative artefact. Given the superior near-field resolution of ultrasound and the superficial position of the lesion, it would have been a better choice of imaging in the first instance.

In Case 2, the role of intraoral ultrasound was not as an alternative imaging modality to MRI but rather a mechanism of choosing the most appropriate imaging pathway. In this role, the convenience, patient tolerance and relative financial cost of ultrasound (in comparison to CT and MRI) cannot be overlooked, especially when concern is being raised about MRI waiting times.¹⁵ Whilst it is unlikely that ultrasound equipment will be employed in general dental practice in the near future, cost-benefit and risk-benefit (no

ionizing radiation) considerations already justify its use in hospital for thyroid and salivary gland imaging.

In conclusion, the sonographic appearances of these lesions corroborated the histopathological behaviour, specifically in distinguishing benign or low grade, from aggressive. In addition, the case outcomes demonstrate the convenience and effectiveness of intraoral ultrasound as a prequel or possible alternative to MR or CT in assessing intraoral soft tissue lumps for excisional or incisional biopsy. Custom-designed intraoral probes facilitate its “chairside” use as an extension of clinical examination. These technical developments are applauded and wherever possible, the use of intraoral ultrasound as a tool in oral medicine is positively encouraged.

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Consent

Written informed consent for the case to be published (including images, case history and data) was obtained from the patients for publication of this case report, including accompanying images.

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