CASE REPORT

Mucoepidermoid carcinoma mimicking a mucocele (ranula) in the floor of the mouth

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Mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy. The submandibular gland is rarely involved, about less than 10% of all other major salivary glands. We report a case of a 49-year-old female who presented with a mass on the left side of the floor of the mouth. She was complaining of neck pain for the last 2–3 days and had noticed the mass growing in size and becoming tender to palpation. There was no exacerbation of pain during eating, drinking, or opening of mouth. A multidetector CT scan was made. The radiographic examination revealed a mostly well-defined hypodense area suggestive of mucocele (ranula), which was thought to be corroborated by the clinical findings. The treatment of choice was the complete excision of the mucocele and the left submandibular salivary gland. The histopathological examination surprisingly revealed a low-grade MEC. No further treatment was recommended; however, a follow up was scheduled in 4 months. This atypical location of MEC in the floor of the mouth simulated the clinical and radiographic features of a mucocele. This case highlights the value and importance of using all the available diagnostic tools and modalities for optimal patient management, as the histopathological analysis played an important role to confirm the diagnosis.

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Introduction

In general, parotid gland tumours are benign while neoplasms of the sublingual, submandibular and minor salivary glands are more likely to be malignant.^{1,2} Among the salivary gland tumours, mucoepidermoid carcinoma (MEC) is the most common malignancy.^{3,4} Approximately, more than half of these tumours are present in the major salivary glands; the parotid gland being the most frequent location. The others occur in the minor

salivary glands, primarily at the palate. The submandibular gland is rarely involved, about less than 10% of all other major salivary glands. This tumour develops over a wide age range—from second to seventh decades of life. Histologically, the MEC is classified depending on its aggressiveness into three grades: low, intermediate, or high grade.^{4,5} Rarely, it may be present in the jaws as an intraosseous tumour, called central MEC.^{4–6}

Although variable, the clinical presentation of MEC of minor salivary glands often takes on a benign appearance of a soft tissue mass. Since tumour cells present the ability to produce mucin and form cystic spaces, some of the MEC can resemble ranulas or other mucoceles,

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presenting as red-purple, fluctuant, smooth-surfaced masses.⁴ Ranula is a term used for mucoceles (mucus escape reactions or mucus extravasation phenomenon) that occur in the floor of the mouth, arising from the sublingual gland. Other similar swellings in the floor of the mouth (*i.e.* salivary duct cysts, dermoid cysts and cystic hygromas) may be erroneously classified as ranulas.

We report an atypical location of MEC in the floor of the mouth simulating the clinical and radiographic features of a mucocele. This case also highlights the value and importance for optimal patient management of the histopathological analysis even of very otherwise benign-looking lesions.

Case report

A 49-year-old female presented to the University of Iowa Hospital and Clinics with an intraoral, soft, fluctuating, cystic mass in the left side of the floor of the mouth. The patient described having ongoing symptoms for 5 months prior to her appointment at the hospital. She complained of neck pain for the last 2–3 days and had noticed the mass growing in size, moving posteriorly in her neck and becoming tender to palpation. She did not notice any exacerbation of pain during eating, drinking, or opening of the mouth. Clinical examination showed no external signs of infection or inflammation, and intraorally, no signs of tooth-related infection could be seen. Past medical history was non-contributory. At that visit, the patient presented with results of a previous whole-body positron emission tomography/CT (PET-CT) scan and fine needle aspiration (FNA) biopsy of the intraoral lesion, both performed at an outside facility. According to the PET-CT scan report, a 3 cm, rounded, centrally

necrotic/cystic mass with a rim of mild hypermetabolic uptake along its periphery in the left submandibular space was observed with no other remarkable findings (Figure 1). No lymphadenopathy was noticed and all major organs (lungs, heart, liver, spleen, kidneys and bladder) were unremarkable. The FNA biopsy report stated the presence of few epithelial cells, which made the diagnosis inconclusive. Based on the previously reported findings and the current clinical observations, a multidetector CT (MDCT) scan was made, revealing a low-density cystic mass measuring approximately 2.6 cm with a slightly irregular/thickened peripheral border (Figure 2). The borders were mostly well-circumscribed with no inflammatory changes in the adjacent fat, and no suspicious lymph nodes or destructive bony lesions were noticed. A complete excision of the lesion, followed by excision of the left submandibular salivary gland, was performed.

The biopsied specimen, on gross examination, was represented by a tan-pink, intact, encapsulated soft tissue nodule, measuring $3.4 \times 2.9 \times 2.9$ cm (Figure 3). Sectioning revealed a unilocular cyst filled with red-brown, thin fluid with a tan-pink, webbed to undulating lining with patchy tan-yellow discolouration. The left submandibular gland, also excised, appeared as a tan-pink to yellow, partially encapsulated soft tissue, measuring $4.7 \times 2.9 \times 1.7$ cm and containing a possible intraparenchymal lymph node. No other discrete lesions or masses were identified.

Histologic examination surprisingly revealed a low-grade MEC with classic features. The tumour was intermixed within a soft tissue matrix composed of solid and prominent cystic isles of epidermoid epithelium, intermediate cells and mucous cells (Figure 4). The tumour measured 3.4 cm in greatest dimension and no lymphovascular space or perineural invasion was identified. All surgical margins were free of tumour. The

Figure 1 FDG PET-CT axial (a) and coronal (b) slices showing a rim of mild hypermetabolic uptake (arrows) along the periphery of a cystic mass in the left submandibular salivary gland region. FDG PET-CT, fluodeoxyglucose positron emission tomography/CT.

Figure 2 MDCT axial (a, d), coronal (b, e) and sagittal (c, f) slices in both bone (top row) and soft tissue (bottom row) windows demonstrating a low-density cystic mass with a slightly irregular/thickened peripheral border (arrows). MDCT, multidetector CT.

excised submandibular gland showed unremarkable salivary gland tissue and the presence of a benign reactive intraglandular lymph node.

After the biopsy and unexpected diagnosis of low grade MEC, the patient had returned for a routine follow up session showing no symptoms other than mild residual tenderness at the anterior portion of the incision. On examination, the skin scar was healing well, there was no fluid collection and no mass by bimanual examination. The floor of the mouth was intact with

no damage to tongue motion or sensation. Since the tumour was small, localized and with a low grade histological appearance, no further treatment was recommended. Moreover, all margins were free of tumour, no extraparenchymal extension and no lymph node involvement was present. The patient will return for a 4-month follow up after the excisional biopsy and a new CT will be performed in 6–12 months.

Discussion

Based on the clinical and radiographic findings, the main differential diagnosis included plunging ranula, mucocele and cystic hygroma. Ranula is a clinical term used for mucoceles that occur in the floor of the mouth, arising from the sublingual gland. A simple ranula remains within the sublingual space while a diving or plunging ranula extends to the submandibular and/or parapharyngeal spaces.^{7–9} On CT, ranulas present as homogeneous unilocular low-density lesions with fine walls, unless secondarily infected, or after surgery.⁷ Mucoceles are mucous-filled cavities that are present as soft, fluctuant, painless swellings.¹⁰ Since mucoceles originating from the submandibular gland are extremely rare, a diagnosis of plunging ranula was at first favoured in this case. However, plunging ranulas have a pathognomonic sign represented by a small longitudinal tail-like extension of the pseudocyst into the sublingual space

Figure 3 View of the gross surgical specimen.

Figure 4 Low (1×) and high (10×) power, haematoxylin- and eosin-stained, slides showing a predominately cystic tumour with focal solid areas of epidermoid epithelium, intermediate cells and mucous cells. The findings were consistent with a low-grade mucoepidermoid carcinoma.

that could not be seen in the CT scan of the present case, making the radiographic diagnosis challenging. On the other hand, cystic hygroma rarely presents in the submandibular space, but if present, it reveals similar images on CT scan to those of a plunging ranula. Cystic hygroma is infiltrative in nature and typically involves contiguous anatomic regions in the neck. It appears well-defined but poorly circumscribed, lobulated, unilocular or multilocular, homogeneous and with fluid attenuation on CT images. Differentiation of cystic hygromas from ranulas is based on their typical lack of association with the sublingual space and by its potential for multilocularity.⁷ The present case presented as a well-circumscribed lesion with no signs of lobulation or septation, thus not favouring cystic hygroma. Surgical excision is the first-choice of treatment for both mucocele and cystic hygroma. The removal of the involved gland to reduce the chances of recurrence is recommended in cases of mucocoeles.

Although clinical and radiographic aspects, as well as the non-aggressive behaviour of the lesion, favoured the diagnosis of mucocele, a salivary gland neoplasm that may present with a cystic component should also be included in the differential diagnosis for the present case. Moreover, as FDG PET-CT has been reported not to be helpful in differentiating benign from malignant salivary lesions,^{11,12} one should be cautious when interpreting mild hypermetabolic uptake along salivary glands, as seen in the present case.

With its frequent cystic presentation and the most common salivary gland malignancy, MEC should be considered. MECs are classified depending on their histological aggressiveness using a 3-tier system of grading: low, intermediate, or high grade.^{4,5} A combination of cystic and solid patterns and the presence of mucous, epidermoid, intermediate, columnar and clear cells can be seen in many cases and helps to determine the diagnosis. The present case showed the tumour tissue present in only a small area in the wall of mainly a cystic lesion. This may indicate that the original lesions were in fact retention cysts, in which part of the ductal epithelium of the cyst wall changed to a MEC.¹³

In this present case, given that the FNA pathological report described rare presence of epithelial cells and no solid component, and the clinical and radiographic benign appearance of the lesion, we decided to treat it with an excisional biopsy including the excision of the left submandibular salivary gland. The literature supports complete excision to confirm diagnosis and decrease morbidity and mortality^{2,14,15} and reports that radical surgery is contraindicated in small, localized with low- to intermediate-grade.³ Radiotherapy should only be used for patients with unclear margins and positive lymph nodes,⁵ which was not the case in the present report. Besides that, low grade MEC presents excellent prognosis if treated early² emphasizing how important is to provide a correct and early diagnosis to the patient.

The present case showed a low-grade MEC mimicking a ranula in the floor of the mouth. The benign clinical and radiographic features lead to a more conservative treatment with no suspicion of malignancy. Since MEC can resemble cystic and inflammatory lesions, clinicians must be aware of the importance of a differential diagnosis and, also that the histopathology is mandatory for all cases. Although not very helpful in this case, the FNA biopsy represents a good first diagnostic tool and may provide a preliminary assessment of the lesion or result in a definitive diagnosis. When inconclusive results are obtained from FNA, incisional or excisional biopsies are keys to determine the diagnosis and guide the treatment. Overall, the present case highlights the value and importance of using all the available diagnostic tools and modalities for optimal patient management, as the histopathological analysis played an important role to confirm the diagnosis and redefining the treatment plan and follow up accordingly.

Consent

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

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