Case Report

Difficulties in Diagnosing Lesions in the Floor of the Mouth – Report of Two Rare Cases

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Abstract

This article highlights 2 contrasting lesions of the floor of the mouth, the first being a benign lipoma growth and the latter, an adenoid cystic carcinoma. Both of these lesions appear clinically similar, presenting as a swelling with normal overlying mucosa and otherwise asymptomatic at the time of clinical examination. As the swelling for Case 1 is small and fluctuant, no special investigation was ordered, whereas a computed tomographic scan was ordered for the larger expansile lesion in Case 2. The lesions were excised under local and general anaesthesia respectively and a histology henceforth. Recovery for both cases were uneventful and no recurrence or complication was noted to date when this article was written. The two extreme natures of the lesions manifested in the region serve as a cautionary note to clinicians.

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Introduction

The floor of the mouth takes form as early as in the fourth week of gestation with the downward growth and subsequent degeneration of the ectoderm surrounding the peripheral of the tongue, forming the lingual sulcus separating the tongue and the floor of the mouth. The anterior boundary is demarcated in the sixth week with the formation of the Merkel's cartilage, which later ossifies to form the mandible. At about this point in the fetal development, focal thickenings appear in the oral epithelium. These foci proliferate into the underlying ectomesenchyme to form buds which then morphogenise and cytodifferentiate to form the salivary gland. The formation of these structures entwine with a network of nerve, blood vessels and lymphatic system forming the bulk of the floor of the mouth.

It is also important to learn from the embryological development of the neutral plate that the floor of the mouth was once upon a time closely related to the mediastinum. This association is evident even after gestation where layers of neck muscles bridge these 2 anatomical spaces. This is a significant clinical association as infection originating from the floor of the mouth has the potential to tract along these muscle planes down to the vital structures in the mediastinum.

Therefore, any lesions arising from the floor of the mouth are tightly related to the confined structures present in this anatomical space except thyroglossal tract cyst, which has a different embryological origin. Table 1 tabulates a list of possible lesions and corresponding structures arising from the floor of the mouth. However, these predictable structures in this confined space do not ease the diagnosis. In fact, lesions in this region often manifest with a swelling that mimics another clinically. This could be attributed to the bulk of soft tissues which help mask the lesional appearance, especially the deeply seated ones. This article illustrates 2 such cases.

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Case 1

A 55-year-old Chinese man presented with a fluctuant swelling on the right floor of the mouth, which was incidentally discovered by a general dental practitioner

Table 1. Floor of the Mouth Lesions

Inflammatory/trauma	Mucus retention phenomenon (Ranula)
Developmental	Dermoid cyst Epidermoid cyst Lymphoepithelial cyst
Neoplasm	Lipoma Salivary gland tumour Benign mesenchymal tumours

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during routine dental check up. As the lesion was asymptomatic, the patient was unaware of its presence prior to the diagnosis. He was subsequently referred to an Oral Maxillofacial Surgeon for management.

At rest, the lesion resembled a bulk of excess soft tissue at the floor of the mouth herniating through an edentulous ridge on the lower right quadrant. The lesion became more obvious when the patient raised his tongue. Bimanual palpation delineated a fluctuant mass measuring approximately 15 mm in diameter. The patient presented to the Oral and Maxillofacial Surgery Clinic in National University Hospital in October 2000. From the clinical signs and symptoms, the diagnosis made was a ranula. However, the possibility of the ranula having a plunging phenomenon could not be excluded.

Local anaesthesia marsupialisation of the lesion was performed in October 2000. Incidentally, incision and dissection via the mucosa directly over the crest of the mass yielded an unexpectedly soft, yellowish lobulated mass (Fig. 1). The classical sight of an adipose tissue coupled with a capsule surrounding its peripheral reclassified our diagnosis as a lipoma. Excision of the lesion was performed instead and careful dissection performed to segregate the lipoma from the contiguous sublingual gland. The lesion measured 20 x 14 x 10 mm and was isolated in total. Apart from the sublingual gland, other associated structures, such as the submandibular ducts and lingual nerve, were not exposed intraoperatively.

A portion of the lesion was separated and displaced into water to differentiate its relative density and the rest of the specimen was sent for histopathology. Buoyancy of the lesion in water reaffirmed the presence of fatty tissue (Fig. 2).

Histopathological examination revealed a benign tumour composed of several lobules of adipose tissue (mature fat cells). This was consistence with the diagnosis of a lipoma.

Healing was uneventful and a 3-year review follow-up yield was also nil.

Case 2

A 54-year-old lady attended the Oral and Maxillofacial Surgery Clinic in May 2002 with a complaint of a large lump below the left side of her tongue for about 4 months' duration (Fig. 3). A course of antibiotics had been prescribed by her doctor and she was referred for further investigations and management. Her speech and masticatory efficiency were affected by the swelling. The lesion was, however, asymptomatic and there appeared to be no neurosensory deficit. It was firm and non-tender on palpation and measured about 4 cm clinically. The differential diagnosis made included ranula, dermoid cyst and salivary gland tumour.

A computed tomography (CT) scan was performed and the findings indicated a slightly heterogeneous soft tissue mass measuring 1.8 x 3.6 cm in its largest cross section in the anterior floor of the mouth on the left (Fig. 4). No significant lymphadenopathy was noted.

Excision of the left sublingual mass and the associated sublingual gland was performed under general anaesthesia. The submandibular duct was cannulated and the lingual nerve protected (Fig. 5a). The mass and sublingual gland were excised and sent for histopathological examination (Fig. 5b). Her postoperative recovery was uneventful.

Biopsy of the mass revealed adenoid cystic carcinoma with characteristic cribriform epithelial nests forming cylindromatous growth pattern in a fibrous stroma. Occasional solid mass was also noted. No neural invasion was identified and the tumour appeared intact within the capsule. Biopsy of the sublingual gland was negative for any malignancy.

The case was discussed at the multi-disciplinary Tumour Board and decision was made to give the patient radiation therapy (a total of 60 Gy over 30 fractions was given) in view of the high risk of local relapse. The patient has since been on regular follow-up with no complications. Clinical and radiographic examinations are being done regularly with no evidence of recurrence.

Discussion

Floor of the mouth lesions can be very challenging and the common diagnosis of a ranula for floor of mouth swellings can be misleading, as shown by our 2 case reports.

Lipoma is a slow growing benign tumour and intra-oral incidence ranges from as low as 1% to as high as 4.4% in the other studies.[1-3] Lipids unavailable for metabolism[4] coupled with the autonomous growth of a lipoma have rendered it to be a true benign neoplasm.[5] The cheek is the commonest site of occurrence in the intra-oral cavity followed by the tongue, floor of the mouth, buccal sulcus and vestibule, palate, lips and gingival.[6] This pattern corresponds closely to the quantity of fat deposits in the intra-oral cavity. The slow expansile rate of a lipoma probably allows neighbouring structures to adapt to the increasing pressure and leaves the patient oblivious of the growth. A case of denture instability attributed to the growth of a lipoma has been reported.[7] Another case of unremarkable history but remarkable swelling in the floor of the mouth was reported by Gray and Barker in 1991.[8] Though oral lipoma rarely exceed 25 mm, the case reported in this article would have taken the same fate as reported in the above literature had the dentist attending to the patient had not been vigilant enough to notice the swelling in his dental examination.

Morphologically, intra-oral lipomas can be grouped into

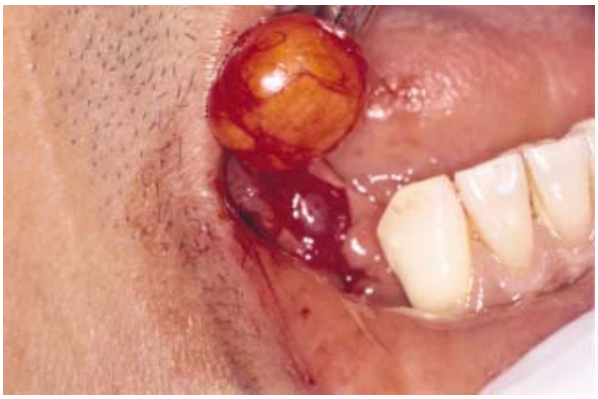

Fig. 1. Intraoperative appearance of the lipoma removed.

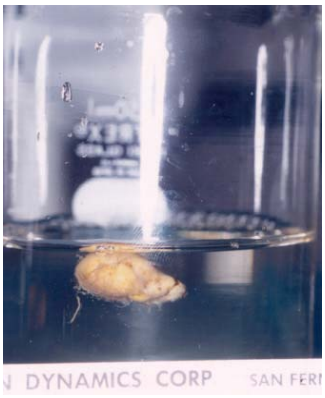

Fig. 2. Hypodensity of lipoma leads to buoyancy in water.

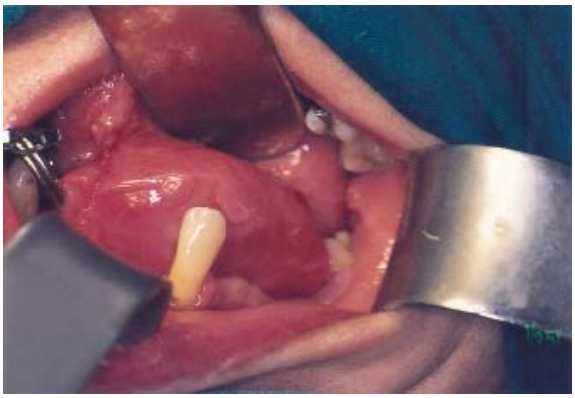

Fig. 3. Clinical appearance of the adenoid cystic carcinoma in the floor of the mouth.

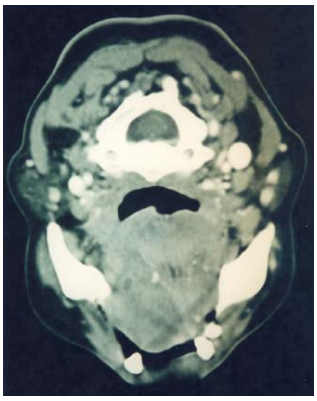

Fig. 4. Computed tomographic illustration of the adenoid cystic carcinoma lesion in the floor of the mouth.

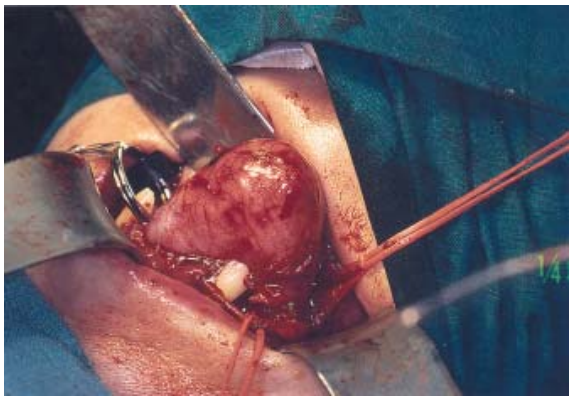

Fig. 5a. Intraoperative appearance of the adenoid cystic carcinoma.

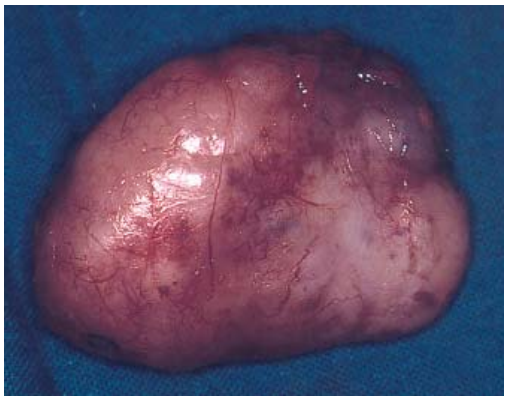

Fig. 5b. The excised adenoid cystic carcinoma.

2 forms: a diffuse form affecting the deep tissues and a superficial, encapsulated form.[8] The former group may warrant some caution surgically as more vital structures are entailed in the operation.

At this point, it may also be prudent to draw attention to a possible phenomenon of an infiltrating lipoma. Unlike the normal lipomas, infiltrating lipomas are not encapsulated and invade readily into the deep soft tissue.[9] This accounts for 62.5% of its recurrence after surgical excision.[10]

Histologically, lipoma comprises of mature fat cells. It is, however, interesting to note the existence of spindle cell lipoma as first reported by Enzinger and Weiss in 1975.[11] Spindle cell lipoma occurs 1 in 60 of the incidence of the ordinary lipoma.[12] An important significance of this lesion is its ability to transform to a liposarcoma or a fibrosarcoma.[12]

CT, ultrasonography and magnetic resonance imaging (MRI) are useful adjuvants in the investigation of lesions of the floor of the mouth. Specific range of CT Housefield

unit values has been tabulated for lipomas. In ultrasonographic pictures, lipoma often appears hypoechoic.[13] MRI's superiority in soft tissue contrast enables a lipoma to be isolated and its multi-planar images are particularly useful in observing infiltration into the oral floor.[13]

The aetiology of lipoma remains a mystery. A list of possible aetiologies is shown in Table 2.[2,14-16]

Table 2. Possible Aetiologies of Lipoma

1. Lipoblastic embryonic cell nest in origin[2]
2. Metaphase of muscle cells[2]
3. Fatty degeneration[2]
4. Hereditary[14]
5. Hormonal cause[14]
6. Trauma induced[15,16]
7. Infection[15,16]
8. Chronic irritation[15,16]
9. Infarction[15,16]

Lipomas are treated by surgical excision and do not yield recurrences[17] (non-infiltrating type) due to the well-defined demarcation by the capsule. However, surgical manipulation of lipoma in the floor of the mouth must be cautioned as the lesion maybe in contiguous with vital structures such as the salivary glands and their respective ducts and the lingual nerve. Manipulation or iatrogenic trauma to the salivary glands or ducts may result in the formation of a ranula.

Adenoid cystic carcinoma or cylindroma,[18] on the other hand, is the second most common malignant epithelial tumour of the salivary glands and comprises about 4% to 8% of all salivary gland tumours. Fifty per cent to 70% of all reported cases occur in minor salivary glands of the head and neck. It is also the most common malignant tumour of the submandibular gland.[19,20]

This tumour has a slightly higher incidence in females with peak incidence in the 5th to 7th decade. It commonly presents as a slow-growing enlarging mass with severe pain in 50% of cases and occasionally with facial nerve paralysis (25%) and neurosensory deficit especially in late-stage lesions.[21,22] This tumour can also invade through bone marrow.

Adenoid cystic carcinoma has a history of relentless recurrences, systemic spread and becoming progressively more aggressive after surgical resection.[23] It is one of the most biologically deceptive and frustrating tumours in the head and neck region. This is accounted for histologically by its infiltrative capacity and distinct propensity for perineural invasion.[24,25] It has been shown that brain-derived neurotrophic factor (BDNF) is a growth factor known to be involved in neurogenesis. BDNF is uniformly expressed by adenoid cystic carcinoma and may play a causative role in its predilection for perineural invasion.

The origin of adenoid cystic carcinoma is thought to be from the intercalated duct reserve cell or terminal tube complex. There are 3 histological types of adenoid cystic carcinoma, but all exhibit peripheral nerve invasion. The first type is the cribriform type, with classic histology of nests of cells with neatly punched out spaces. Cystic-like spaces are not true ductal or glandular lumina but are in continuity with the supporting connective tissue stroma. The second type is the tubular type, in which the tumour cells are arranged in small nests separated from one another. There are true duct lumens surrounded by differentiated ductal cells and islands of tumour cells lie in hyalinised stroma. The third type is the solid type with variable sized rounded or lobulated aggregates of tumour cells with few cyst-like spaces.[26]

There is no strong correlation between histologic type and behaviour of the tumour, but some argue that the tubular and cribriform types have a better prognosis.[27,28] Size, anatomical location, facial nerve involvement and presence of metastases are the most important prognostic factors, including gross residual tumour after surgery.

Blood-borne metastases in 40% to 60% are common with the lung being the recipient in 40% of cases. Bone and liver are other sites of tumour metastasis. Lymph node metastases are uncommon unlike other salivary gland carcinomas. Wide to radical excision with postoperative radiation therapy is the treatment of choice.[29]

Adenoid cystic carcinomas are generally considered to be resistant to conventional forms of radiotherapy (photons/X-rays or electrons) because of their ability to repair radiation damage. However, in the setting of low disease burden after surgery, conventional radiotherapy is effective in reducing the risk of local tumour recurrence. Neutron radiotherapy is now being used. Neutrons are an entirely different type of radiation which have different radiobiological properties than conventional radiation. A neutron is a constituent of the atomic nucleus and interacts directly with the nuclei in tissue. Compared to conventional radiation, neutrons typically deposit 20 to 100 times more energy along their path length and so inflict greater damage in the cells, which is less readily repaired. Neutron radiotherapy has been tested on many different types of tumours but salivary gland tumours and, in particular, adenoid cystic carcinomas are where neutrons show the greatest benefit. Neutrons deposit greater amounts of energy in all tissues through which they pass, not just the tumour cells, giving rise to more severe side effects than conventional radiotherapy. Hence, neutron radiotherapy is generally utilised for patients with inoperable or recurrent disease or when there are multiple positive margins (indicating a considerable amount of residual disease), if

there is perineural invasion noted in the operative specimen, or if there is tumour spread to the regional lymph nodes. It can also be used in cases where there is recurrence in areas that have been previously treated with conventional radiotherapy. Such cases must be evaluated individually as the side effects associated with an aggressive re-treatment of this type tend to be severe. In the case of tumours located in the head and neck region, this generally means giving 16 treatments over a 4-week period (conventional radiotherapy generally takes 7 to 8 weeks). If the tumour is located elsewhere in the body, it may be necessary to give the treatment over a longer time.

Once distant metastasis has occurred, about 20% of patients may survive from 5 to 14 years. However, 33% of patients usually die within less than 2 years after distant metastases. Overall, the 5-year survival rate is 80% to 90%. At 15 years, the rate is only 10%.

Conclusion

Floor of the mouth lesions remains challenging clinically and surgically. The common occurrences of superficial ranula and sialoliths often simplify the possible diagnosis in the floor of the mouth. However, it has been illustrated in the literature and the cases reported in this article that lesions in this anatomical position could pose conflicting diagnosis. As lesions in this region range from the most benign ranula to the sinister carcinoma, it may be prudent to investigate further with CT, MRI or ultrasonography. Perhaps a further study of all the lesions of the floor of the mouth and its incidence is necessary to justify these investigations.

Deeply seated lesions of the floor of the mouth also pose possible surgical complications. Tightly netted vital structures in contiguity with each other and its possible resultant infection, which could tract down to the mediastinum, are morbidities surgeons fears most.

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