Abstract

Pleomorphic adenoma, also known as benign mixed tumor, is the most common benign tumor of salivary glands that mostly occurs in the parotid or submandibular glands, but may also be found in the minor salivary glands that are distributed throughout the oral cavity. Surgical removal with adequate margins is the principal treatment. Due to its microscopic projections, this tumor requires a wide resection to avoid recurrence. We report a case of a pleomorphic adenoma in the upper lip, the second site for frequency for benign tumors of minor salivary glands, after the hard and soft palate.

Key words: minor salivary glands, pleomorphic adenoma, oral cavity

INTRODUCTION

Tumors of the minor salivary glands represent 10-15% of all salivary gland neoplasms (1). An estimation of the incidence of salivary gland neoplasms in the population, is that, for every 100 tumors of the parotid gland, there are likely to be 10 of the submandibular gland, one of the sublingual gland, and 10 of the minor salivary glands. The probability of a malignant diagnosis is less than 25% in patients with a tumor of the parotid gland, about 50% in those with a tumor of the submandibular gland, more than 80% in patients with a tumor of minor salivary gland origin, and virtually 100% in those few with a tumor of the sublingual gland (2).

Benign tumors of minor salivary gland origin are most frequently pleomorphic adenoma and have been located in areas as diverse as tongue (3), posterior part of the tongue (4), nasal cavity and septum (5), larynx (6), and trachea (7). The most common site for pleomorphic adenoma is the hard palate, followed by the upper lip. Some of these pleomorphic adenomas can become massive with malignant degeneration before presentation (8).

CASE REPORT

A 57 year-old female presented with a painless submucosal swelling in the right half of the upper lip, with a history of 1 year, in the Department of Oral and Maxillofacial Surgery Iași, in May 2010. Her past medical history was uneventful and there was no previous history of regional trauma. On clinical examination there was a 2 cm/1.5 cm firm, painless, circumscribed lesion in the right half of the upper lip, adherent to the underlying structures and the mucosal surface was non-ulcerated (figure 1). There was no regional lymphadenopathy and her general condition was normal. A clinical differential diagnosis of squamous cell carcinoma with nodular outset and a tumor of minor salivary gland origin (benign or malignant) were considered. The entire tumor was excised with a wide margin, on local anaesthesia (figures 2, 3). The result of histopathological exam was: pleomorphic adenoma in the upper lip, with a particularity: it microscopically simulated a myoepithelial carcinoma (figure 4). There was no recurrence at three month follow-up.

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Fig. 1. Painless non-ulcerative, submucosal swelling in the upper lip, right half

Fig. 2. Intraoperative view of dissection in benign tumor of minor salivary gland originating in the upper lip

Fig. 3. The operative specimen

Fig. 4. The histopathological result: pleomorphic adenoma in the upper lip (particularity: bilayered duct-like structures with conspicuous outer layer of clear myoepithelial cells simulating a myoepithelial carcinoma)

DISCUSSION

Minor salivary glands, coming up to 450-1,000 in number, are widely distributed in the head and neck area (9). In their majority (70-90%), they are located in the oral cavity and oropharynx, including the lateral margins of the tongue, lips and buccal mucosa, palate, glossopharyngeal area, and the retromolar trigone. The remaining ones are located in the nose, paranasal sinuses, pharynx and larynx. Minor salivary glands contribute to about 8-10% of the volume of whole saliva.

The signs and symptoms of tumors associated with minor salivary glands vary according to their different anatomical sites. The majority of patients were 60 years of age or older. Many of the larger series had reported a gender distribution of 66% women. The most frequent site of origin is the oral cavity and oropharynx and, within the oral cavity, most tumors develop in the region of the hard palate because this is the area with the highest density of minor salivary glands. Most of patients present a painless non-ulcerative, submucosal swelling. The mucosal layer is adherent to the mass and a small ulcer may be present during its evolution. Up to 26% of patients presented local pain (1).
Physical examination and awareness that a clinically "benign" submucosal swelling at any place in the head and neck may be a tumor of minor salivary gland origin and that, statistically, the pathology of that tumor is more likely to be malignant rather than benign, is the most important clinical information that will improve accurate diagnosis, and allow for a rational plan of management of these tumors.

Currently, imaging using computed tomography and/or magnetic resonance imaging may help to the delineation of the tumor, an accurate staging of the disease, and also for a correct planning of a surgical procedure.

The use of fine-needle aspiration cytology (FNAC) in tumors of minor salivary gland origin may be helpful in correctly classifying the tumor as benign or malignant, however, the use of incisional biopsy or punch-biopsy may create a better and more representative specimen, thus revealing the correct histological type.

The current incidence of a benign tumor of salivary gland origin is estimated to be 60-80/ million people per year. The incidence of malignant salivary tumors in the United States of America is 10/1 million people per year and in the United Kingdom is 0.6/1 million per year (10).

Benign tumors of minor salivary gland origin are most frequently pleomorphic adenoma and the most common sites are the hard palate and the upper lip. Other benign neoplasms of minor salivary gland origin have been reported in the oral cavity (11). Benign epithelial tumors also include: Warthin tumor, monomorphic adenoma, intraductal papilloma, oncocytoma, and sebaceous neoplasms. Benign nonepithelial tumors (mesenchimal origin) include: hemangioma, angioma, lymphangioma (cystic hygroma), lipoma, and neural sheath tumors.

Microscopically, there is a variety of configurations of pleomorphic adenomas, but the essential components are: capsule (complete or incomplete), epithelial cells in a variety of configurations, myoepithelial cells, stroma (which may be fibrocollagenous, mucoid, chondroid or myxochondroid, in varying proportions, but which form the bulk of the tumor in the majority of cases) (11). We presented in this paper a rare case of a pleomorphic adenoma in the upper lip, which microscopically looks like a myoepithelial carcinoma.

Surgical excision is the treatment of choice for benign tumors of minor salivary glands in the oral cavity. The treatment will depend on the size and location of the tumor. Recurrence depends upon the accuracy of local excision, and most patients with recurrence will have it within 18 months. Long-term follow-up is recommended, as the risk of recurrence may remain life long for such patients (12).

CONCLUSIONS

- Tumors of minor salivary gland origin are uncommon and are more likely to be histologically malignant than benign.
- Clinical presentation for a benign tumor of minor salivary gland origin in the oral cavity is most commonly a non-ulcerative, submucosal swelling.
- Incisonal or punch-biopsy should be performed prior to planning radiological imaging and/or final treatment.
- The variety of microscopic configurations of pleomorphic adenomas of minor salivary glands is remarkable, but stroma represents the bulk of the tumor in the majority of cases.
- Complete surgical excision is the preferred treatment.
- Pleomorphic adenoma remains the most common benign neoplasm for minor salivary glands and the treatment is local excision with a safe margin.

References


