ORIGINAL ARTICLE

Histology of Submandibular Gland Tumours, 10 Years’ Experience

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KEYWORDS
Neoplasm; Submandibular gland; Benign; Malignant; Pleomorphic adenoma

Abstract
Introduction and objectives: There are few reports focused on neoplasms in the submandibular gland because they are uncommon and are frequently grouped with the other salivary glands tumours. In the classical studies, the proportion of malignancy reported in these cases is around 50%.
Objectives: Determining the proportion of malignancy, the most frequent histological types, the gender distribution and average age at diagnosis in patients with submandibular gland neoplasms who were treated in our hospital from 2000 year to 2010.
Methods: A retrospective review of our department database of the patients who underwent surgery for submandibular gland neoplasm in a ten-year period was performed.
Results: Twenty-two patient records were included, in which 19 (86%) patients presented benign disease and 3 were of a malignant type (adenocarcinoma, well-differentiated epidermoid carcinoma, follicular lymphoma). Two of the 3 cases of malignant neoplasms were in men.
Conclusions: A greater proportion of benign neoplasm was found in submandibular tumours. The most frequent benign tumour is the pleomorphic adenoma. Women are more commonly affected (76%). Benign tumours appear in younger patients than do malignant ones.

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PALABRAS CLAVE
Neoplasia; Glándula submandibular; Benigna; Maligna; Adenoma pleomorfo

Histología de tumores de glándulas submandibulares. Experiencia de 10 años

Resumen
Introducción y objetivos: Hay pocos trabajos enfocados a neoplasias de glándulas submandibulares porque son poco comunes y se les agrupa con otras glándulas salivales. En los estudios clásicos se reporta que los tumores malignos en esta localización representan alrededor del 50%.

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**Introduction**

Salivary gland tumours comprise 3% of head and neck tumours. The overall incidence of these neoplasms is 4/100,000 per year, with the gender ratio being 1:1. The average age of onset is 45 years, with a peak between the fifth and sixth decades of life. The mean age is higher for carcinomas than for adenomas. The most frequently affected gland is the parotid, representing between 64% and 80% of cases, followed by the submandibular gland with 7%-15%, and finally the minor and sublingual salivary glands with 1%-5%. According to the classical studies by Simons and Spiro carried out in 1964 and 1976 with a total of 345 patients, benign neoplasms constitute 75%-80% in the parotid, 50%-60% in the submandibular gland and 35% in the minor and sublingual salivary glands; the smaller the affected salivary gland, the greater the likelihood of malignancy.

The most frequent neoplasms in the submandibular glands are: pleomorphic adenoma (which represents 36% of cases), adenoid cystic carcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumour (10%). Clinical reports indicate that benign neoplasms are characterised by a painless enlargement of the submandibular triangle. Malignant neoplasms are clinically manifested by a more rapid increase in volume, coupled with the presence of pain and neurological involvement due to involvement of the marginal branch of the facial nerve.

Tumours originating in the major salivary glands are accessible to biopsy by fine needle aspiration (FNA). An experienced cytopathologist can reliably distinguish malignant salivary pathologies from benign, but a histological classification based on only aspiration is an unrealistic goal. Computerised tomography and magnetic resonance imaging enable accurate assessment of tumour extension, compression or infiltration of adjacent structures, presence of nodal metastases and better planning of the therapeutic approach. The definitive diagnosis is done through a histopathological study.

There are currently few reports focused on submandibular gland neoplasms. This is because they are rare and are usually grouped with other salivary glands. Most studies relating to frequency and prognosis of these neoplasms are from centres specialising in cancer or pathology departments. Little is known about the frequency of submandibular tumours treated at non-specialised centres. The proportion of malignant tumours of the submandibular gland varies from 10% to 78.2% in different reports; this in turn depends on several factors, among which are race and location where the information is collected, since tertiary referral centres find a higher proportion of malignant tumours. Most classical studies report malignancy rates greater than 50% in submandibular gland tumours. However, more recent studies in different populations (Asian, Brazilian) have reported smaller incidences, with a malignancy rate of 10%-30%.

In this study, we report the prevalence of different submandibular gland neoplasms treated at our hospital during the period from 2000 to 2010.

**Methods**

We reviewed the records of cases registered in the database of the Otolaryngology Division at Dr. Manuel Gea González General Hospital from January 2000 to June 2010, who had undergone surgery due to submandibular gland neoplasm and in which there was a histopathology report. The variables analysed were age, gender, family history, consumption of tobacco and alcohol, type of neoplasm and histological diagnosis.

**Results**

In the period from 2000 to 2010, there were a total of 22 records of patients with submandibular gland neoplasms. Neoplasms of benign type were predominant, representing 86% of the total. The age range was 13–66 years. The mean age of cases with benign pathology was 40 years, whilst the mean age of malignant cases was 55 years. The majority appeared in female patients (76%), although 2 of the 3 cases of malignancy occurred in males.

Of all patients with benign neoplasms, 47% had a history of smoking and 26% of alcohol consumption. Of all benign neoplasms, the most frequent was pleomorphic adenoma, which represented 89% of the total.
Pleomorphic adenoma appeared more frequently in female patients (76%). Of these patients, 47% had a history of smoking; 23% referred alcohol intake and 11% (2 cases) reported a family history of salivary gland neoplasm, but were unable to clarify the histology.

Malignant neoplasms encountered were adenocarcinoma, well-differentiated squamous cell carcinoma and follicular lymphoma, with 1 case each.

In total, 2/3 of patients with malignant tumours were smokers and all of them consumed alcohol.

Warthin’s tumour appeared in one male patient with a history of smoking and drinking (Table 1).

**Discussion**

We found that 86% of submandibular gland neoplasms were benign and 14% were malignant. This data differs from that in classical studies of submandibular gland tumours; nevertheless, it agrees with recent studies performed with a smaller number of cases.1-10

In our literature review, we found that the global distribution of these neoplasms varies.13-16 In a 10-year retrospective study on an Asian population of 101 patients in 2010, Denis et al. reported a low incidence of malignant tumours in submandibular glands (21.1%).7 In a 5-year retrospective study in Singapore on 93 patients from a tertiary centre, Goh et al. found that 13.5% of submandibular tumours were malignant,8 which is consistent with the trend in Asian populations with a low prevalence of malignant tumours in the submandibular gland.

The most common benign neoplasm in our series was pleomorphic adenoma, whilst malignant neoplasms found were adenocarcinoma, well-differentiated squamous cell carcinoma and follicular lymphoma. It is noteworthy that we did not find any cases of adenoid cystic carcinoma, the most common malignancy in this location in other series. However, having only 3 cases to study represents a constraint in drawing conclusions in this regard. In a 10-year retrospective study carried out in a UK general hospital, Ethunandan et al. identified 25 patients with primary submandibular gland neoplasm, of which 68% were benign and all cases were of pleomorphic adenoma.9 Malignant tumours represented 32% and adenoid cystic carcinoma was the most frequent histological subtype, followed by carcinoma ex pleomorphic adenoma, mucoepidermoid carcinoma and salivary duct carcinoma. The frequency of histological subtypes in this series was similar to that in previous reports in the literature.11

In a Brazilian population study carried out in 2009, De Oliveira found that malignant tumours accounted for 19% of submandibular gland neoplasms and that adenoid cystic carcinoma was the most frequent. However, there are reports that mucoepidermoid carcinoma is the most frequent. Adenocarcinoma was the second most common, but it is reported as the third in frequency in other series. The age distribution in this study ranged from 1 to 88 years, with a mean age of 45 years and a peak incidence around 30-39 years. The mean age for malignant tumours (55 years) was significantly higher than for benign tumours (43 years).10 In our series, the mean age at the time of diagnosis of benign tumours (40.7 years) was lower than the mean age at diagnosis of malignant neoplasms (55.3 years). The age range in
our patients with pleomorphic adenoma was 13–65 years, with a predominance of females over males.

De Oliveira reports that salivary gland tumours affect females more often, with a male:female ratio of 1:1.5. This ratio is reported as 1:1.6 in benign tumours and 1:1.5 in malignant tumours. Similarly, other studies describe a male predominance in a ratio of 1:4:1. In our cases, benign neoplasms occurred in a male:female ratio of 1:2.8, whilst malignant cases presented a ratio of 2:1. The case of Warthin's tumour is particularly striking, as it is a benign neoplasm that appears almost exclusively in the parotid gland.

The subjects who underwent surgical excision of the tumour commonly described an asymptomatic growth that remained stable for years and sought medical attention when there was a greater increase in volume or accompanying symptoms appeared.

Population studies conducted in different geographical areas can provide more information as to the most common types of neoplasm and their clinical behaviour. Such studies can also generate new lines of research for this diverse group of tumours.

Conclusions

In agreement with findings from other recent series, benign tumours in our study represented a major proportion of submandibular gland neoplasms. Pleomorphic adenoma was the most common neoplasm in the submandibular gland.

In our series, submandibular neoplasms occurred more frequently in females. Submandibular gland neoplasms generally appeared around 40 years of age when they were benign and around 55 years when they were malignant.

Conflict of Interests

The authors have no conflicts of interest to declare.

References