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(CASE REPORT)

Pleomorphic adenoma of the sublingual gland: A case report

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Abstract

Pleomorphic adenoma, also known as mixed tumor, is the most common neoplasm arising from salivary glands, accounting for approximately 81.2% of all benign tumors in salivary glands. The parotid gland accounts for 85% of these tumors, with 10% arise in the submandibular gland and only 0.3% in the sublingual gland. The aim of this study is to describe the specific management of pleomorphic adenoma in its unusual location. Here, we present a case of pleomorphic adenoma of the sublingual gland in a 62-year-old woman, manifesting with a painless mass in the floor of the mouth for 5 years, appearing as a well-limited hypodense tissue lesion, enhanced after contrast injection, in the sublingual region and extending to the right mylohyoid muscle in computed tomography imaging. The tumor and sublingual gland were resected under general anesthesia. The histopathological diagnosis was pleomorphic adenoma. No evidence of recurrence was detected after three years follow-up.

Keywords: Pleomorphic Adenoma; Salivary Gland Neoplasm; Sublingual Gland

1. Introduction

Salivary gland tumors are rare and account for 3.5-10% of all head and neck tumors. Tumors of the sublingual salivary glands accounting for 0.3-5.2% of all epithelial salivary gland tumors and approximately 1.5% of all carcinomas of the main salivary glands [1, 2]. Most tumors arising from the sublingual gland are malignant, with an estimated prevalence of 80-90% [3, 4]. In contrast, benign tumors such as pleomorphic adenoma (PA) are rarely reported [5-6]. Due to the rarity of PA in the sublingual gland, most observations from various reports lack detailed comparisons of pathological findings and long-term outcomes. Here, we present our own case and provide a comprehensive review and discussion of the existing literature.

2. Case report

A 62-year-old woman with no medical history presented with a painless mass in the right floor of the mouth that had been present for 5 years. Examination of the oral cavity revealed a well-bounded, mobile, firm, lobulated mass of the right buccal floor covered with normal oral mucosa, measuring 4 cm x 3 cm. The mass was not attached to surrounding structures, including the mandible (Figure 1). Saliva flow from the right submandibular canal was undisturbed. The patient had poor dental condition with no signs of local infection, tongue mobility and taste were normal. There was no facial asymmetry or cervical adenopathy, and the rest of the ENT examination was unremarkable.

The computed tomography scan revealed a relatively well-defined, hypodense tumor, enhanced following contrast injection, in the sublingual region and extending to the right mylohyoid muscle. The tumor was measured at 43.5x45x26.5mm (Figure 2).

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The tumor was excised using an intra-oral approach under general anesthesia. A longitudinal incision was made in the mucosa of the floor of the mouth opposite the tumor, which was then dissected with the sublingual gland into a single mass minutely from the muscular and vascular structure of the floor of the mouth (Figure 3). The tumor did not adhere to the surrounding tissue.

The surgical procedure was successful in maintaining hemostasis, and no intraoperative or postoperative complications were reported. These included the absence of hematoma of the floor of the mouth and no paralysis of the tongue or taste problem.

Histological examination revealed the presence of a salivary parenchyma containing an encapsulated tumor proliferation, which consisted of two distinct contingents: an epithelial and a stromal component. The proliferation is organized into lobules, which are separated by a hyaline and discreetly myxoid stroma. This is compatible with a diagnosis of pleomorphic adenoma (Figure 4).

The clinical and CT scan follow-up was unremarkable, with no evidence of tumor recurrence after three years (figure 5).

Figure 1 Right buccal floor mass covered by intact oral mucosa

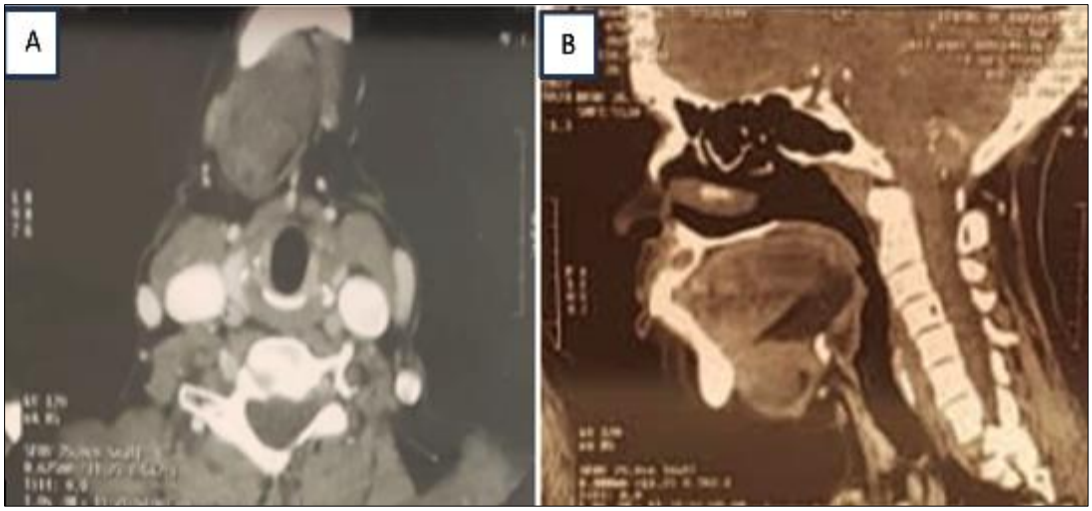

Figure 2 CT scan with Axial (A) and Sagittal (B) cuts showing a fairly well-limited, hypodense tumor of the sublingual gland extending to the right mylohyoid muscle

Figure 3 Surgical excision piece

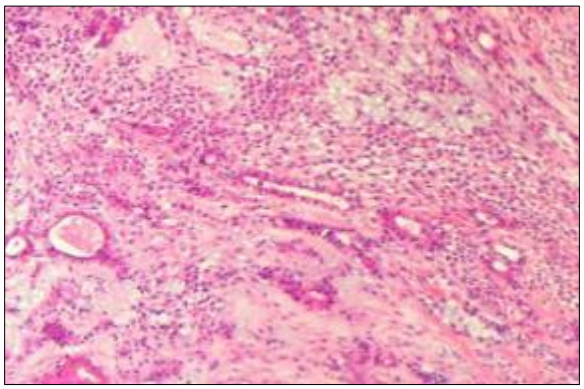

Figure 4 Tumor with a dual epithelial and mesenchymal composition, with a myxoid stroma (magnification X 100, Hematein Eosin stain)

Figure 5 Photo of the buccal floor one year after surgery showing the absence of tumor recurrence

3. Discussion

Salivary gland tumors account for only 3.5-10% of head and neck tumors, and tumors of the sublingual glands are the least common compared with tumors of the parotid, submandibular and minor salivary glands [7]. The majority of these tumors are malignant, and a few benign cases of pleomorphic adenomas have been described in the sublingual glands [6-8]. Therefore, when a tumor of the sublingual glands is suspected, as in the present case, it is essential to take into account the high probability of malignancy.

PA is the most common type of salivary gland tumor, with a prevalence of 81.2%. The sites most susceptible to tumorigenesis are the parotid glands (85%), followed by the submandibular glands (6-11%), the minor salivary glands

(10-23%) and the sublingual glands (0.3-2%) [1-9]. PA is typically slightly more common in women, and age of onset is more widely distributed than in other salivary gland tumors [5]. Clinically, an asymptomatic swelling in the floor of the mouth is the most common complaint. Tumors of the sublingual salivary gland are generally only recognized at an advanced stage, mainly because of their minimal symptomatology. In terms of disease duration, tumor growth tends to be slow, and subjective symptoms such as ulcers, pain, and functional impairment are rare, so it tends to go untreated for relatively long periods.

Imaging is essential in the preoperative assessment of PA. On CT and MR images, PA present as well-defined lesions occasionally accompanied by characteristic lobulated contours. On T2 weighted images, typical PA show marked hyperintensity, which reflects the abundant myxochondroid stroma, with a hypointense rim indicating the fibrous capsule. However, intratumorally signal intensity varies according to the cellular density, proportion of epithelial and stromal components, and type of stromal components [10].

Incision biopsy of large salivary gland tumors is often criticized due to the risks of tumor cell dissemination and nerve damage and the procedural difficulties posed by tumor adhesion [11,12]. For these reasons, fine-needle aspiration biopsy (FNAB) is frequently performed because it is a simple and minimally invasive procedure [13]. However, FNAB has low diagnostic accuracy for salivary gland tumors due to the variety of histological features and multitude of histological types, and samples are poor due to the low cell sampling count [13,15].

Early total surgical resection of the sublingual gland and its neoplastic mass in normal margins for benign tumors is the treatment of choice to avoid recurrences. The excision may possibly be accompanied by selective neck dissection and radiotherapy for malignancies. The histologic examination of the lesions is necessary not only to establish the diagnosis but also for the better management of the surrounding tissues [15]. Depending on the tumor size and location, the total resection may become quite complex due to the close proximity of the sublingual gland to the inner cortex of the mandible, the submandibular salivary gland and its duct, the lingual vessels and nerve, and the hypoglossal nerve [8, 15]. In our case, clinical and CT imaging features indicated a close-to-surface, circumscribed, easily accessible tumor, and thus an intraoral total resection of the lesion was performed.

Pleomorphic adenomas are generally benign tumors. It can recur as well as it can give rise to a malignant transformation. Long-term clinical and radiological monitoring, lasting for years, is essential after to watch out for these complications.

4. Conclusion

The pleomorphic adenoma is a heterogeneous benign tumor combining various epithelial and mesenchymal structures epithelial and mesenchymal structures. It is less common in the accessory salivary glands, particularly the sublingual gland. Because of the risk of local recurrence and malignant degeneration, all patients undergoing surgery for a pleomorphic adenoma must undergo regular clinical monitoring.

Compliance with ethical standards

Disclosure of conflict of interest

There are no conflicts of interest to declare related to this research

Statement of ethical approval

While formal ethical approval was not obtained for this study, we ensured that all aspects of the research were conducted ethically and with respect for the rights and well-being of the participants.

Statement of informed consent

Informed consent was obtained from all participants involved in the study, and this information has been appropriately included in the manuscript.

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