Pleomorphic Adenoma of the Submandibular Gland: A Case Report

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Pleomorphic adenoma is the most common benign tumor of the salivary glands. About 80-90% of pleomorphic adenoma occurs in the parotid gland and about 5-10% of them occur in the submandibular gland and sublingual gland. The rest proportion comprises the minor salivary glands. The palate is the most common area, followed by the lips and the cheeks. Pleomorphic adenoma of the submandibular gland shows clinically a slow-growing, painless mass found on submandibular aspect. In this case report, we report a case of pleomorphic adenoma of the right submandibular gland in a 65-year old female patient who was referred from the department of neurosurgery, due to the heterogenous mass detected by computed tomography (CT) and magnetic resonance imaging (MRI).

Key words: Pleomorphic adenoma, Submandibular gland, Salivary gland tumor

I. INTRODUCTION

Salivary gland tumors are rare and take up 3% of head and neck tumors.1) Pleomorphic adenoma is the most common salivary gland tumor, accounting for about 40-70% of all major and minor salivary gland tumors. The most common site of this tumor in minor salivary gland is the palatal area followed by the lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, and retromolar area.2) As the name of pleomorphic adenoma suggests this tumor have mixed histology and consists of 3 components: An epithelial, a myoepithelial and a stromal (mesenchymal) component. It is also known as benign mixed tumor which describes its pleomorphic appearance on light microscopy having mixed origin from epithelial and myoepithelial elements.3)

These well circumscribed tumors with ductal and myoepithelial elements are found in both the major and minor salivary glands with most occurring in the parotid gland.1) Approximately 90% of the benign neoplasm of the major salivary gland is associated with the parotid gland, Pleomorphic adenoma comprises 80-90% of these benign parotid neoplasms. Pleomorphic adenoma of the submandibular and sublingual gland is uncommon (8-10%)5) Pleomorphic adenoma of the minor salivary gland is comprises rest of the group. The most common sites for pleomorphic adenoma of the minor salivary glands are the palate (42.6%), with
equal incidence in both the hard and soft palate, followed by the lips (10.1%) and the cheeks (5.5%).

The most frequent neoplasms in the submandibular glands are: pleomorphic adenoma (36%), adenoid cystic carcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumor (10%). Clinical reports indicate that benign neoplasms are characterized by a painless enlargement of the submandibular triangle.

Pleomorphic adenoma may occur at any age, but the highest incidence is in the fourth to sixth decade of life. Forty percent of them are male, 60% female. The risk of the pleomorphic adenoma becoming malignant is about 6%. Pleomorphic adenoma usually presents as a slow-growing, painless mass, which may be present for many years. Symptoms and signs depend on the location. The treatment of choice for pleomorphic adenoma of submandibular gland is total submandibular gland excision along with tumor.

In this case report, we report a pleomorphic adenoma of the right submandibular gland in a 63-year old female patient who was referred from the department of neurosurgery, due to the heterogenous mass detected by computed tomography (CT) and magnetic resonance imaging (MRI).

II. CASE REPORT

1. Diagnostic Work-up

A 63 years old female was referred from department of neurosurgery at Seoul St, Mary’s Hospital of the Catholic University of Korea to the department of oral and maxillofacial surgery at same hospital for evaluation of the heterogenous mass detected by CT and MRI. She was being treated by neurosurgeons for trigeminal neuralgia. Medical history, dental history and family history were not contributory.

In the extra-oral examination, there was no abnormality, No abnormal findings were observed in the lymph nodes, And, also no abnormal findings were observed in the intra-oral examination.

In the radiographic examination, we evaluated two images of CT and MRI. In enhanced neck CT, there was a round low density less enhancing mass at the superior portion of the Rt. submandibular gland, 1.9 × 2.5 × 1.5 cm, (Fig. 1, 2) In enhanced neck MRI, there was an about 1.8 × 2.3 × 1.6 cm sized T2 high signal intensity, gradually enhancing lesion in Rt. parapharyngeal space. It was tight abutting superior portion of submandibular gland and closely abutting medial pterygoid muscle. (Fig. 3, 4) On the basis of the radiographic examination, a provisional diagnosis was made as salivary gland tumor arising from right submandibular gland or hemangioma by radiologist. Due to the low prevalence of hemangiomas in adults with about 50 cases reported worldwide, we did not taken into consideration in the differential diagnosis of salivary gland masses.

We decided to diagnose salivary gland tumor arising from right submandibular gland provisionally and performed surgical excision for final diagnosis.

2. Treatment and Outcome

After preoperative investigations, we planned for surgical excision. The patient was operated under general anesthesia, A 4 cm incision in placed in lateral neck crease approximately 2-3 cm below the lower edge of the mandible, Subplatysmal skin flap was developed and the marginal mandibular nerve
was identified and protected. The facial vein was identified and ligated at the inferior border of the gland and reflected superiorly with the fascia over the submandibular gland. This maneuver exposed the submandibular gland and ensured protection of the marginal mandibular nerve. The facial artery was ligated and preserved by ligating only the branches of the facial artery to the gland. Blunt dissection then continued towards the superiomedial gland at which point the mylohyoid muscle was retracted anteriorly to complete the dissection. Posterior and inferior traction on the gland facilitated identification and differentiating Wharton's duct, the lingual nerve with its attachment to the submandibular ganglion, and the hypoglossal nerve. The submandibular duct was then ligated and divided close to its opening in the floor of the mouth. The gland was liberated from the submandibular ganglion and removed preserving the lingual and hypoglossal nerves. We inserted the drain to prevent edema and hematoma and layered suture was done. After the operation daily dressing was done for 3 days. Drain was removed 3 days after surgery and stich out was performed 5 days after surgery. There was no complication and no damage of marginal nerve, hypoglossal nerve, lingual nerve. There was no recurrence after a follow-up period of 6 months.

The excised mass was sent for histopathology study (Fig. 7, 8). After 1 week, the histological result showed a small solid tumor in the right submandibular gland. The tumor was well encapsulated with fibrous bundles. The tumor cells...
were mostly composed of myoepithelial cells forming myxoid stroma and epithelial sheath, accompanied with ductal structures. There was no tumor necrosis or malignant transformation, (Fig. 9, 10) This tumor was benign and well localized at the glandular tissue. The definitive histopathology report confirmed the diagnosis as pleomorphic adenoma, early stage of right submandibular gland.

Fig. 5, Operative view during surgical excision

Fig. 6, Drain insertion

Fig. 7, Gross appearance of the excised specimen (1)

Fig. 8, Gross appearance of the excised specimen (2)

Fig. 9, Photomicrograph

Fig. 10, Photomicrograph
III. DISCUSSION

Salivary gland tumors comprise 3% of head and neck tumors. 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. Most salivary gland tumors are benign, with malignancy found in roughly 14% of lesions. Pleomorphic adenoma is the most common benign tumor of salivary gland, while mucoepidermoid carcinoma is the most common malignant counterpart to be encountered in maxillofacial region.

Majority of the salivary gland tumors affect parotid gland with more than 70% of the cases. Several studies have been conducted on the tumors of the parotid and minor salivary glands, but very few reports in the literature have focused on submandibular gland tumors as they are rare and are usually grouped with other salivary glands. Submandibular gland is affected in 5-10% of the cases with pleomorphic adenoma being the most common tumor.

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 tumor (10%). Clinical reports indicate that benign neoplasms are characterized 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.

Pleomorphic adenoma is the most common salivary gland tumor, accounting for about 40-70 % of all major and minor salivary gland tumors. These well circumscribed tumors with ductal and myoepithelial elements are found in both the major and minor salivary glands with most. They are more common in women and age at diagnosis is mostly between 40 and 59 years old.

A neck mass can be a diagnostic challenge in patients of any age and is commonly evaluated with CT, MRI or ultrasound. CT and MRI can help in determining the location, size of tumor and extension to the surrounding areas specially bone. MRI can provide a better information of the vertical and inferior tumor extension and it more accurately indicated the degree of encapsulation. The diagnosis of pleomorphic adenoma is based on complete and thorough history, physical examination, cytology and histopathology. Fine needle aspiration findings provide evidence for a pre-operative diagnosis that is 70-80% accurate and also helps to differentiate between tumor and inflammatory conditions or enlarged lymph nodes. The final pathologic diagnosis is always established based on findings from surgical excision.

Apart from conventional methods of diagnosis newer technique like Immunohistochemistry (IHC) can be used to help differentiate pleomorphic adenoma from other tumors. The IHC stains that have proved useful are Keratin Cam 5.2 and EMA P-63 Calponin, Maspin, S-100 HHF-35 Muscle-specific actin Glial fibrillary acidic protein BMP and Aggrecan.

In this case, radiologist made a provisional diagnosis as salivary gland tumor arising from right submandibular gland or hemangioma on the basis of the radiographic examination. But, due to the low prevalence of hemangiomas in adults, we decided to diagnose salivary gland tumor arising from right submandibular gland provisionally and performed surgical excision for final diagnosis.

In general, the complete resection of tumor and
submandibular gland is recommended due to the high rate of local recurrence in this disease. The excision of the tumor should also be accompanied by the removal of the submandibular gland in toto. Historically, simple enucleation is associated with unacceptably high recurrence rates of up to 45%. Pleomorphic adenoma generally does not recur after adequate removal and shows a high cure rate of more than 95%. The local recurrence is thought to be mainly caused by the rupture of the tumor capsule during the operation or the misunderstanding of the tumor margin. Incomplete removal of the glandular tissue paves the way for a definitive recurrence. Therefore, the enucleation of tumor is thought to be avoided. Recurrence rate of submandibular gland tumors are less than parotid gland since entire gland is excised. Pleomorphic adenoma is benign tumors with a well-documented transformation to malignancy (carcinoma ex pleomorphic adenoma). It is estimated that up to 25% of untreated PAs undergo malignant transformation. Therefore, early definitive treatment is strongly recommended.

In this case report, we report a case of pleomorphic adenoma of the right submandibular gland in a 63-year old female patient who was referred from the department of neurosurgery, due to the heterogenous mass detected by CT and MRI. We decided to diagnose salivary gland tumor arising from right submandibular gland provisionally and performed surgical excision immediately. Surgery was performed under general anesthesia and excised both the submandibular gland and the tumor at the same time. There was no complication and no damage of nerve. There was no recurrence after a follow-up period of 6 months. Long-term follow-up will be needed in the future because of the risk of recurrence.

REFERENCES


