Case Report

Symptomatic pleomorphic adenoma of the submandibular gland in a male patient: A case report and review of the literature

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ABSTRACT

Salivary gland tumors represent only 1–4% of head-and-neck tumors, with submandibular gland tumors representing around 10% of salivary gland tumors. Although rarely seen, submandibular pleomorphic adenoma is the most common of the submandibular gland neoplasms (36%). These tumors have a female predominance and present as a painless mass. We report a 42-year-old male patient with a 6-month history of swelling in his left submandibular region due to submandibular pleomorphic adenoma in a hospital in North India. Many diseases in the submandibular region with overlapping clinical pictures make diagnosing difficult. Integrated clinical, radiological, and cytological approaches help in differential diagnosis and making a provisional diagnosis. However, the final diagnosis can be established only by histopathology.

Key words: Otolaryngology, Pleomorphic adenoma, Salivary gland tumors

Salivary gland tumors are rare and comprise only 1–4% of head-and-neck tumors, and most of the tumors of the salivary glands arise from the parotid gland (70%), whereas submandibular gland tumors represent about 5–10% of salivary gland tumors [1,2]. Tumors of the minor salivary glands have a higher risk of being malignant, about 60–80% [3]. Of the tumors affecting the salivary glands, the most common is pleomorphic adenoma or benign mixed salivary gland tumor, seen most commonly in the fourth to seventh decade of life with a female predilection [4]. Most pleomorphic adenomas (80%) arise from the parotid, while those from the submandibular gland are lesser (12%). Thus, the submandibular pleomorphic adenoma is considered a rare tumor [5,6]. Pleomorphic adenoma has a heterogeneous histology and a possibility of getting malignant. The malignant transformation rate for pleomorphic adenoma varies from 1.9% to 23.3% [5]. In the case of incomplete surgical excision, the pleomorphic adenoma can recur due to pseudopods and incomplete capsules. Therefore, a complete excision of the tumor is needed [7]. Many studies have been done on parotid gland tumors, but only a few reports have focused on tumors arising from submandibular glands, as they are rare.

In this paper, we describe a pleomorphic adenoma that arises from the submandibular gland of an adult patient of male gender and correlate its clinical, radiological, fine needle aspiration and histopathological findings that established the diagnosis of pleomorphic adenoma.

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CASE PRESENTATION

A 42-year-old male patient came with a 6-month history of swelling in his left submandibular region. The swelling was asymptomatic initially and of small size; however, it increased gradually with time, causing mild dysphagia, and reaching the size with which the patient presented to us (Fig. 1). The patient was a smoker. Medical history and personal history were not contributory.

On extraoral examination, there was a diffuse swelling in the left submandibular region measuring about 6 cm × 5 cm in size. On palpation, the swelling was firm, non-tender, non-fluctuant, and mobile with well-defined borders. Facial and neck muscular movements were regular, and dental caries were present on intraoral examination.

The patient was advised an initial ultrasound (USG) of the neck that revealed a 57 mm × 34 mm hypoechoic lesion seen in the left submandibular region just deep and abutting submandibular gland with mild vascularity on Doppler. On the contrast-enhanced computed tomography (CECT) neck, there was evidence of

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a 50 mm × 40 mm hypoenhancing mass lesion seen in the left submandibular region abutting the medial cortex of the mandible but not eroding its wall. The lesion causes a mass effect over the base of the tongue and left lateral wall of the oro-pharynx, protruding inside, and narrowing the oro-pharyngeal lumen. There was no associated significant cervical lymphadenopathy. Scanned parts of bilateral upper lung fields and mediastinum were unremarkable (Fig. 2). Routine blood investigations were within normal limits. Fine-needle aspiration from the swelling revealed myoepithelial and epithelial cells in a typical fibrillary magenta-colored metachromatic stroma, raising the suspicion of submandibular pleomorphic adenoma (Fig. 3). As a result of all of the above findings, a provisional diagnosis of pleomorphic adenoma was made, and it was decided to excise the lesion under general anesthesia surgically.

After informed consent was obtained, the patient was taken for surgery under general anesthesia. Painting and draping were done as usual, with the chin rotated to the other side, and the incision line was marked. An incision about 3–4 cm below the mandible was carried down through the platysma to expose the sternocleidomastoid's anterior border and the digastric muscle's anterior belly. The submandibular gland was identified and mobilized, and the marginal mandibular nerve was preserved. The tumor arising from the submandibular gland was excised, preserving the facial artery and lingual nerve. Wound lavage was done, and hemostasis was achieved. A closed suction drain was put in place, and the wound was closed. The excised specimen was sent for histopathological examination. The post-operative phase of the patient was uneventful, with the use of antibiotics as per hospital policy. The suction drain was removed after 3 days, and skin sutures were removed after 7 days.

The excised mass sent for histopathological examination measured about 6 cm × 5 cm with a bosselated surface. A fibrous capsule surrounded the tumor. On the cut section, it had a grey-white, myxoid appearance. Histopathology of the specimen revealed epithelial cells in ducts and myoepithelial cells in a myxoid stroma, confirming the diagnosis as a pleomorphic adenoma (Fig. 4). The patient is on follow-up and doing well, with no evidence of any recurrence.

Figure 1: Facial view of the patient showing left submandibular swelling

Figure 2: Sagittal and coronal contrast computed tomography sections showing hypoenhancing mass lesion seen in the left submandibular region

DISCUSSION

In our case, the patient was a smoker presenting with a gradual swelling in the left submandibular area that was firm in consistency, and the patient was asymptomatic initially but later presented with dysphagia. USG, computed tomography scan, and magnetic resonance imaging (MRI) as initial investigations

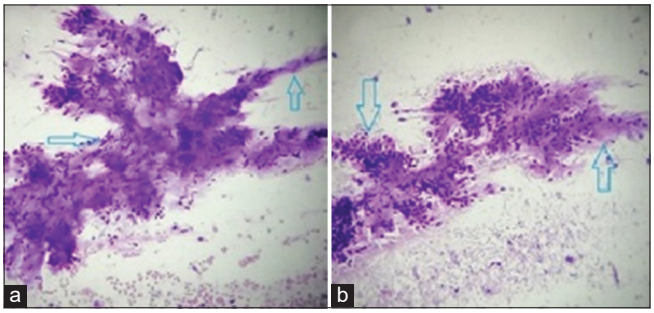

Figure 3: Fine-needle aspiration cytology slides using may Grunwald–Giemsa staining method at ×10 magnification (two different fields a and b). Micrograph showing myoepithelial and epithelial cells in a typical fibrillary magenta-colored metachromatic stroma

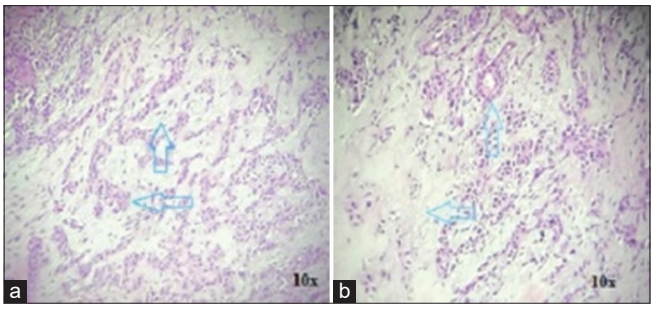

Figure 4: Histopathological slides using H&E staining method at ×10 magnification (two different fields a and b). Micrograph revealing epithelial cells in ducts and myoepithelial cells in a myxoid stroma

can give a reasonable provisional diagnosis of pleomorphic adenoma and show a mass having a well-defined or bosselated border, hyperintense on T2 MRI. Giant tumors commonly have a lobulated delineation that supports the diagnosis [8-10]. MRI aids in determining the perineural invasion and involvement of adjacent soft tissue [7].

Initial USG proved helpful because a hypoechoic lesion was seen in the left submandibular region with mild vascularity on Doppler. Detailed information was obtained by performing a CECT neck that showed a hypoenhancing mass lesion in the left submandibular region abutting the medial cortex of the mandible but not eroding its wall. The lesion was causing a mass effect over the base of the tongue and oropharynx. No associated significant cervical lymphadenopathy, and lung and mediastinum were unremarkable.

Fine-needle aspiration cytology (FNAC) helps in the preoperative diagnosis with about 80% accuracy, differentiating the tumor from inflammatory conditions or lymph nodes [11]. However, histopathology of the surgically excised lesion establishes the final pathological diagnosis. A needle no more than 18G should be used to prevent the seeding of tumor cells in the adjacent areas. In our case, the FNAC revealed the typical fibrillary magenta-colored metachromatic stroma seen in pleomorphic adenoma along with round to oval cells in clusters and singly spread, which raised suspicion of submandibular pleomorphic adenoma.

The treatment of choice for pleomorphic adenoma of the submandibular gland is total surgical excision of the submandibular gland and the tumor [12]. Recurrence is less for submandibular pleomorphic adenoma because the removal of the whole of the submandibular gland is done. Injury to the marginal mandibular nerve is the most common complication due to the compression or stretching of the nerve. The marginal mandibular nerve neurapraxia that occurs in about 25% of cases can be managed conservatively and takes approximately 3 months to recover [13]. Therefore, the incision should be given about 3–4 cm below the lower end of the mandible to avoid nerve injury.

On histopathological examination, grossly, the pleomorphic adenomas may have a cartilaginous appearance and are typically well circumscribed. Microscopically, ductal epithelial cells and myoepithelial cells are proliferated in the mesenchymal stroma. The mesenchymal stroma is an important feature and may be chondroid, myxoid, mucochondroid, hyalinized, osseous, or fatty. Of these, chondroid stroma is specific for pleomorphic adenoma [14].

In the present case, the excised mass had a bosselated surface and a gray-white myxoid cut section. Microscopy revealed that the tumor originated from a mixed salivary gland with a typical pattern of epithelial, myoepithelial cells with stromal components. The stromal component had a myxoid appearance, confirming the diagnosis as pleomorphic adenoma. Since submandibular gland neoplasms are rarely seen and usually grouped with other salivary gland neoplasms, very few reports in the literature have focused on them. Studies that were done previously showed that pleomorphic adenomas of the submandibular and other benign neoplasms of the submandibular region presented with painless swelling of the submandibular triangle and had a female preponderance between the third to fifth decades [1,12]. In our case, the patient was a male in the fifth decade. The patient started with a painless submandibular mass that gradually increased in size and caused dysphagia, for which the patient visited the hospital.

CONCLUSION

Although rare, the diagnosis of pleomorphic adenoma depends on the clinical, radiological, and cytological approaches. The final diagnosis can be established only by histopathology. As the likelihood of submandibular swelling being malignant is high, it is essential to plan the surgery and accurately evaluate the tumor's relation to surrounding structures. A post-operative follow-up of such patients should be done because such tumors can recur and may require radical surgery, although the incidence is very low.

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