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Case Report

Huge Pleomorphic Adenoma of the Submandibular Gland: Report of a Case and Review of Literature

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Abstract

Only a small percentage of salivary gland neoplasms are pleomorphic adenomas, which are uncommon benign tumors of the submandibular gland. The 43-year-old woman in this report had a right submandibular swelling that grew gradually and painlessly over the course of two years. Pleomorphic adenoma was diagnosed by imaging, clinical examination, and fine needle aspiration cytology. The patient's vitals structures were preserved during the full surgical resection of the tumor and submandibular gland. The surgical course was unremarkable, and the diagnosis was confirmed by histopathology. In order to avoid recurrence and possible malignant transformation, this case emphasizes the significance of early detection and complete surgical removal of tumor with the gland.

Keywords: *Adenoma, Pleomorphic, Submandibular Gland Neoplasms, Salivary Gland Neoplasms*

Introduction

Salivary gland tumors are rare, about 3-5% of head and neck neoplasm are covered by this diverse group of diseases[1].The majority of SGT are benign, with 25% coming from minor glands and 70% from major salivary glands[2].Both neoplastic and non neoplastic diseases may develop within any of these sites[3]. The tumors of the parotid and minor salivary glands have been the subject of several investigations, but the submandibular gland tumors have received very little attention in the literature due to their rarity[4]. This paper discusses a huge pleomorphic adenoma submandibular gland and discuss benign salivary gland tumors.

Case Presentation

A 43 year old female came to Oral and Maxillofacial department King Abdullah Hospital Bisha On October 2022. She had a complaint of swelling in right side submandibular area for the last 2 years. According to patient she assumed that the swelling was due to tooth infection and she got extraction of right mandibular premolar, but after extraction the swelling grew in size.

Since that time swelling has been growing slowly. There was no fever or pain related to it and negative for any constitutional symptoms. Previously she didn't get any treatment for this swelling. There was no pus or fluid discharge present. Also, there was no difficulty in swallowing. Past medical history, and systemic history were insignificant.

On general physical examination no signs of anemia, jaundice, cyanosis were present. Extra oral maxillofacial inspection revealed an oval shape swelling present in right side submandibular area. It was 6X5 cm in size roughly. Superior-inferiorly it is extending from the lower border of mandible to hyoid bone level. Posteroanterior swelling is extending from anterior to mastoid and to the hyoid bone. Surface was smooth in appearance [Figure 1]. On palpation a well defined oval shape swelling, non tender, non fluctuant was present. Skin was not attached to under lying swelling. No lymph nodes were palpable. Marginal mandibular and hypoglossal nerve were intact. Intraoral examination and bimanual palpation of floor of mouth, the lesion was mobile. There were no secretions from Wharton's duct. Opposite side submandibular gland was normal. Computed tomogram with contrast was performed to check extent of the lesion. CT scan report revealed 66x52x44 mm well defined mass on axial, coronal and sagittal views. It had calcific foci and homogenous post contrast enhancement. Lesion was occupying carotid space and submandibular space. It was close to carotid bifurcation [Figure 2,3,4]. According to the findings of the history, examination and CT scan, a right submandibular gland tumor was tentatively diagnosed. The patient was advised to have a Fine needle aspiration. FNAC was performed, and the results were compatible with pleomorphic adenoma. Informed consent taken from the patient and surgery was planned under general anesthesia. Right side submandibular incision was used, 2 cm below lower border of mandible [Figure 5]. Blunt dissection carried out and platysma was reached. Platysma was sharply cut with electrocautery. Combination of blunt dissection and diathermy was used and lesion was freed from lower border of mandible, hyoid bone, digastricus [Figure 6].

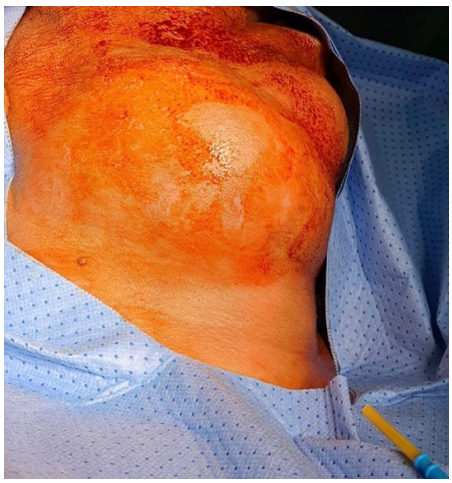

Figure 1. *6x5 cm oval shaped swelling in right submandibular area.*

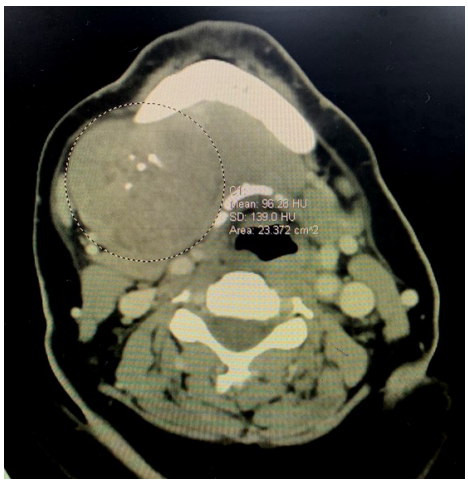

Figure 2. *Computed tomogram axial view showing an iso-dense heterogenous mass in right side carotid and submandibular space. Few calcifications are seen.*

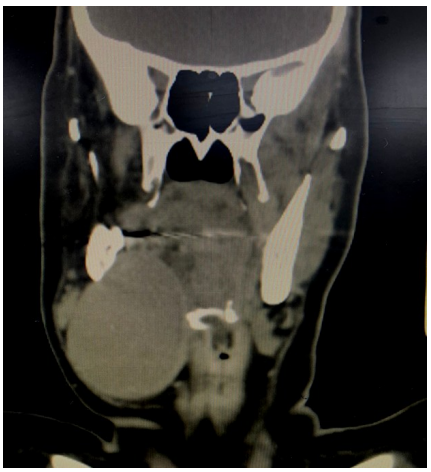

Figure 3. *Coronal view.*

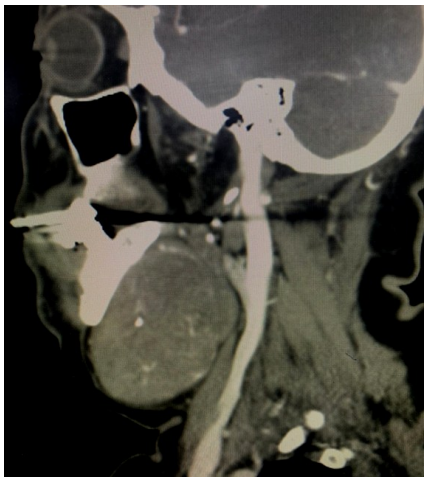

Figure 3. *Sagittal view.*

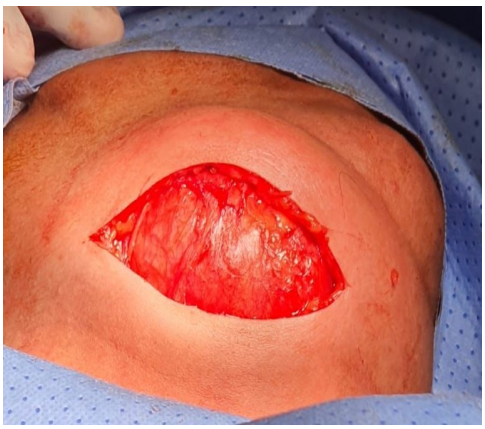

Figure 5. *Right side submandibular incision exposure of platysma muscle.*

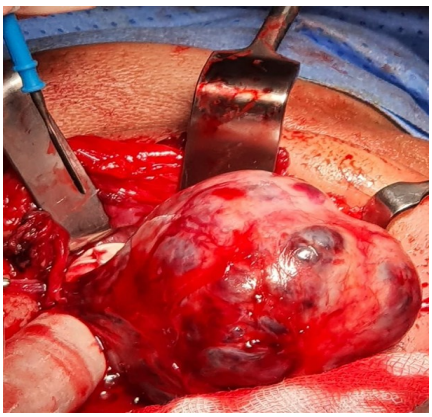

Figure 6.*Intraoperative view , lesion is freed from surrounding structures.*

Dissection was carried out deep to separate it from floor of mouth mylohyoid muscle, carotid vessels. Submandibular duct was ligated; submandibular ganglion was cut and lingual nerve was separated from the lesion. A suspicious lymph node was also removed. Facial artery was ligated with silk 3-0 at 2 sites and facial vein was ligated at one, after that the lesion was removed in a single piece [Figure7, 8]. Hypoglossal nerve, hyoglossus muscle, lingual nerve and mylohyoid muscle was in the view. A 14F silicon vacuumed drain was placed and wound was sutured in layers with 3-0 Vicryl ™ Ethicon. Skin was closed with running subcuticular 4-0 Prolene™ Ethicon sutures [Figure 9]. Tumor was sent for histology. The post-operative course was uneventful; marginal mandibular, lingual hypoglossal nerves were intact and functioning. The suction drain was removed on 2nd day when the serous fluid was less than 10 ml in 12 hours. She was discharged home with oral antibiotics and topical skin ointment. Sutures were removed on 7th post op day. Final histopathology report showed epithelial and myoepithelial cells were seen in chondromyxoid mesenchyme in histopathological sections [Figure 10]. The final diagnosis was PLEOMORPHIC ADENOMA of the submandibular gland. Patient is on regular follow up for any possible recurrence.

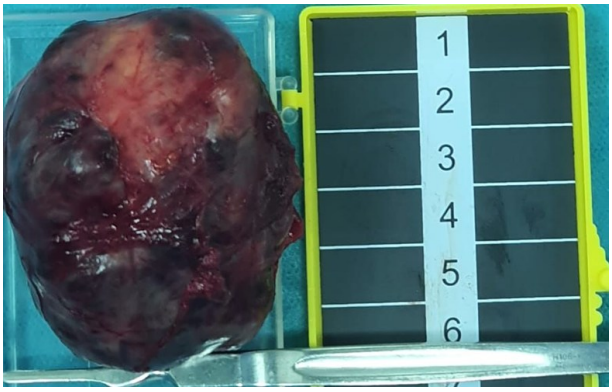

Figure 7. *Removed specimen 6cm height.*

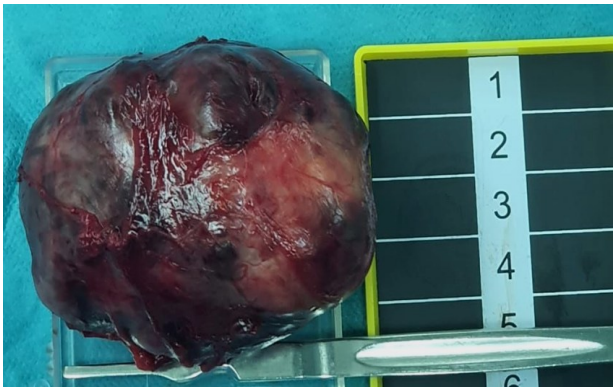

Figure 8. *Removed specimen 5 cm length.*

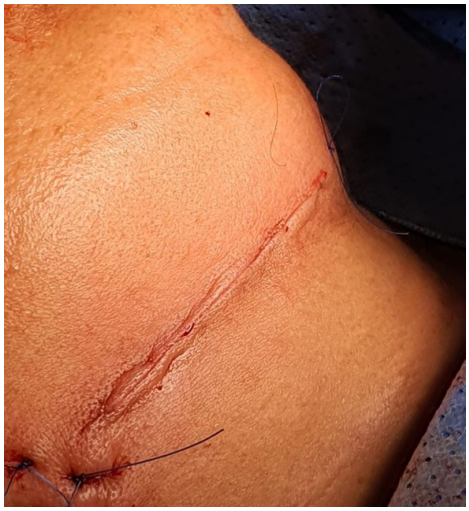

Figure 9. *Skin closure.*

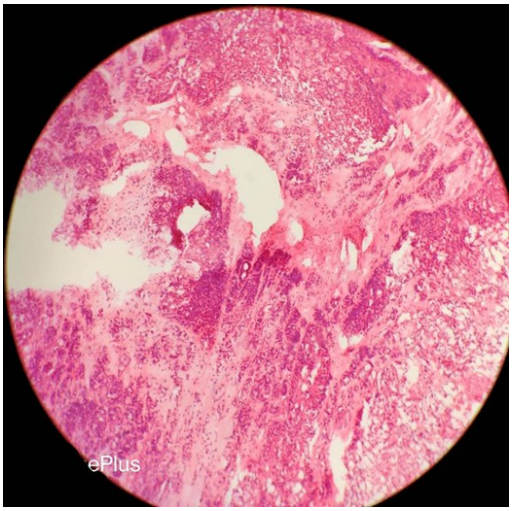

Figure 10. *Histopathology showing epithelial myoepithelial cells with chondromyxoid background.*

Discussion

Salivary gland tumors make about 3-5% of head and neck neoplasms[1]. The most frequent salivary gland tumor is a pleomorphic adenoma[5]. Only 8% of Pleomorphic adenomas originate in the submandibular gland, where 75%–85% of in Parotid gland. The minor salivary glands make the 7–15% of all the cases that have been documented[6]

Alsanie et al. did an international multicentre study on salivary gland tumors. A total of 5739 cases were in the study, 65% were benign and 35% were malignant tumors. For both benign and malignant tumors, there was a small female predisposition (54%), with a peak occurrence between the fourth and seventh decade. The bulk of salivary tumors (68%) manifested in the major glands. The majority of benign tumors were found in parotid gland (70%) and minor glands (47%) had malignant tumors. The most common benign tumors was pleomorphic adenomas (70%) and Warthin's tumors (17%), while the most frequent malignant tumors were mucoepidermoid carcinomas (26%)[7]. Regarding pleomorphic adenoma of submandibular gland, it typically appears as a painless, slowly expanding, movable mass in the submandibular triangle, often neglected by the patient. It does not erode mandibular bone or invade the floor of the mouth. Nerve involvement of lingual, hypoglossal and neck lymphadenopathy are typically absent[8]

Diagnosis is usually made by clinical correlations and by Fine needle aspiration cytology, which can assist in distinguishing between benign and malignant tumors, also inflammatory diseases, enlarged lymph nodes, and other conditions, as well as serve as an evidence for a preoperative diagnosis that is 95.1% accurate[9]. In case diagnosis is not clear, core biopsy can be done[10]. Ultrasound was able to differentiate between benign and malignant lesions with 96% accuracy [11]

Pleomorphic adenomas showed a substantially longer duration to peak intensity and lower maximum curve gradient than the Warthin's tumors in recent methods using contrast enhanced ultrasonography. Warthin tumors typically have higher capillary density rates than pleomorphic adenomas[12]. Usually, a CT scan is enough to detect the extension, but an MRI may be required to view the lesion's capsule. As recommended by Kakimoto et el. [13]

The results of surgical excision are always used to determine the final pathologic diagnosis. Complete removal of the submandibular gland and the tumor is the preferred treatment for submandibular gland pleomorphic adenoma[14]. Preuss et al. Showed that standardised submandibular gland removal is effective treatment with reduced complication rates[15].

Due to the tumor's pseudopod-like extensions, enucleation alone carries a significant risk of recurrence, so tumor should be removed with intact capsule and a small cuff of tissue. The most frequent consequence that causes temporary or permanent paralysis as a result of stretching or compression of the nerve is injury to the marginal mandibular nerve[16]. Regarding the malignant transformation rate there is a recent study shows that among primary pleomorphic adenoma cases, the malignant transformation rate was 11.4%, where as among recurrent cases, it was 20.0%.[17]

Three elements are identified to proper histopathological recognition of pleomorphic adenoma: mesenchymal, myoepithelial, and epithelial. According to histology, pleomorphic adenoma manifests as a variety of epithelial patterns in a loose fibrous stroma of the myxoid, chondroid, or mucoid types. The polygonal cytoplasm of myoepithelial cells is eosinophilic in color[18].

Conclusion

This case highlights the rarity of pleomorphic adenomas occurring in the submandibular gland, as they are most commonly found in the parotid gland. To ensure proper therapy and decrease the chance of malignant transformation, early detection and diagnosis are crucial. Surgical excision remains the main treatment, offering a favorable prognosis in most cases. However, long-term follow-up is vital due to the chances of recurrence, which underscores the importance of careful monitoring postoperatively. The case emphasizes the need for clinicians to consider pleomorphic adenoma in the differential diagnosis of submandibular swellings, particularly when there is a slow-growing, painless swelling, to avoid delays in diagnosis and treatment.

Consent

Informed consent was taken from patient for Clinical Pictures and publication of case for scientific purpose

Conflicts of Interest

The authors declare no conflicts of interest.

References

1. Lawal AO, Adisa AO, Kolude B, Adeyemi BF. Malignant salivary gland tumours of the head and neck region: a single institutions review. Pan Afr Med J. 2015 Feb 12;20:121.
2. Mahomed Y, Meer S. Primary epithelial minor salivary gland tumors in South Africa: a 20-year review. Head Neck Pathol. 2020;14(3):715–723.
3. Kumar MA, Sekhar KC, Kalahasti R. Histopathological study of neoplastic and non-neoplastic lesions of salivary gland: an institutional experience of 5 years. International Journal of Scientific Study. 2017;4(12):69-72.
4. Spiro RH. Salivary neoplasms: Overview of a 35-year experience with 2,807 patients. Head Neck Surg 1986;8:177-84
5. Bokhari MR, Greene J. Pleomorphic Adenoma. 2022 Jul 4. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2022 Jan–. PMID: 28613579.
6. Pinkston JA, Cole P. Incidence rates of salivary gland tumors: results from a population-based study. Otolaryngology–Head and Neck Surgery. 1999 Jun;120(6):834-40.
7. Alsanie, I., Rajab, S., Cottom, H. et al. Distribution and Frequency of Salivary Gland Tumours: An International Multi-center Study. Head and Neck Pathol 16, 1043–1054 (2022).
8. Molina E, Mayer K, Khurana J, Grewal H. Pleomorphic adenoma of the submandibular gland. J Pediatr Surg. 2008;43 (6):1224-1226
9. Allison DB et al. Assessing the diagnostic accuracy for pleomorphic adenoma and Warthin tumor by employing the Milan System for Reporting Salivary Gland Cytopathology: An international, multi-institutional study. Cancer Cytopathol. 2021 Jan;129(1):43-52.
10. Białek EJ, Jakubowski W, Karpińska G. Role of Ultrasonography in Diagnosis and Differentiation of Pleomorphic Adenomas: Work in Progress. Arch Otolaryngol Head Neck Surg. 2003;129(9):929–933.
11. Saito, D Contrast-enhanced ultrasonography for the differential diagnosis of pleomorphic adenomas and Warthin tumors in salivary glands. Laryngoscope investigative otolaryngology, 6(3), 425–431.
12. Kakimoto N et al. CT and MR images of pleomorphic adenoma in major and minor salivary glands. Eur J Radiol. 2009 Mar;69(3):464-72.
13. Eveson JW, Cawson RA. Salivary gland tumours. A review of 2410 cases with particular reference to histological types, site, age and sex distribution. J Pathol1985;146:51-8.
14. Preuss S, Klussmann J, Wittekindt C, Drebber U, Beutner D, Guntinas-Lichius O. Submandibular Gland Excision: 15 Years of Experience. J Oral Maxillofac Surg. 2007;65(5):953-957
15. Hong K, Yang Y. Surgical results of the intraoral removal of the submandibular gland. Otolaryngol Head Neck Surg. 2008;139(4):530-534
16. Song Accuracy of Core Needle Biopsy Versus Fine Needle Aspiration Cytology for Diagnosing Salivary Gland Tumors. Journal of pathology and translational medicine, 49(2), 136–143.
17. Choi et al. Comparative Longitudinal Analysis of Malignant Transformation in Pleomorphic Adenoma and Recurrent Pleomorphic Adenoma. Journal of clinical medicine, 11(7), 1808.
18. Matsumiya-Matsumoto et al. Pleomorphic Adenoma of the Salivary Glands and Epithelial-Mesenchymal Transition. Journal of clinical medicine, 11(14), 4210.