CASE REPORT

Giant Pleomorphic Adenoma of the Submandibular Gland: Case Report and Therapeutic Challenge

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Abstract

Introduction: Pleomorphic adenoma (PA) is the most common benign tumor of major salivary glands.

Case presentation: In this paper, we present a case of PA of submandibular gland of 22 years duration, and it did not undergo malignant transformation. The tumor usually presents as painless, firm, slow growing mobile mass. Fine-needle aspiration cytology was consistent with a pleomorphic adenoma.

Management and prognosis: Surgical excision with preservation of nearby structures was the treatment of choice and post-surgery histopathological examination confirmed the diagnosis of pleomorphic adenoma. After a follow-up period of one year there was no recurrence.

Conclusion: Early diagnosis and prompt treatment is necessary to prevent the risk of recurrence rate. Continuous follow-up of patient is imperative because of the malignant transformation reported in the literature.

Keywords: Pleomorphic adenoma, submandibular gland, mixed tumour.

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Introduction

Salivary gland neoplasms are rare, accounting for less than 2% of all tumors in head and neck.[1-4] Pleomorphic adenoma is the single most common prototypical benign yet true salivary gland tumour. Most of the tumours arise and affect the parotid gland.[1,2] Pleomorphic adenoma of the submandibular gland is an exceedingly rare tumour. Submandibular gland is affected approximately in 5-10% of the cases with pleomorphic adenoma.[3-7] It is usually a benign, slow-growing, well-circumscribed tumour, and is found in all age groups with a predilection for recurrence and risk of malignant transformation. Untreated pleomorphic adenomas may attain a very large size. It is named pleomorphic, or "mixed tumor," because it possesses histologic characteristics of both epithelia and mesenchymal components. We present a case of pleomorphic adenoma of submandibular gland in a elderly woman.

Case Report

A 56-year-old woman presented with a left submandibular mass that had developed over a period of 22 years with complaints of pain for the last 3 months. The mass was enlarging slowly. There was no history of difficulty in swallowing or respiratory discomfort. The patient gave no history of anorexia, weight loss, per-oral bleeding, and passage of stone or change in size of swelling with meals. Routine blood investigations (Hb, RBC, Differential leukocyte count etc) were in the normal range.

Physical examination demonstrated non tender mass measuring approximately 8 cm × 6 cm, normothermic, firm, normal overlying skin, freely mobile swelling was seen involving left submandibular area with no regional lymphadenopathy (Fig. 1).

Ultrasonography (US) with subsequent US-guided fine needle aspiration cytology (FNAC) was carried out. FNA cytology of the cervical mass suggested a mixed tumor of submandibular gland.

The computer-enhanced CT scan of face and neck showed 7.52 cm × 5 cm × 4 cm heterogeneous, well-defined, marginated soft tissue mass in left submandibular area suggestive of PA of the submandibular gland (Fig. 2). The radiographic appearance of tumors in both the locations was suggestive of a benign pathology.

The patient was planned for left-sided submandibular gland excision under general anesthesia and skin incision was given 2 cm below left side mandible. Superior and inferior skin and subplatysmal flaps were raised.

Hence, treatment was complete surgical removal with extraoral approach under general anesthesia preserving marginal mandibular nerve and nearby structures. Perioperative pe-

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riod was uneventful, and the resected specimen weighed 1.07 kg (Fig. 3). No elevation of temperature at the affected site and no pus discharge were seen. The final diagnosis of Pleomorphic Adenoma of right submandibular gland was made. Histopathology showed submandibular gland pleomorphic adenoma composed of myxoid stroma and epithelial cells (Figs. 4A and 4B). Histopathology finding was composed of myxoid stroma and epithelial cells arranged in duct like pattern establishing the diagnosis of pleomrphic adenoma of the submandibular gland.

No correlation was observed between symptom duration and tumor size. Patient was followed-up for a period of 1 year during which there was no recurrence of the tumor and facial nerve functions were normal. Prognosis is excellent except for the rare cases of malignant transformation.

Discussion

Salivary gland tumours are uncommon lesions, and pleomorphic adenoma is the single most common salivary gland tumour. They affect the parotid gland in more than 67% of cases, with the submandibular gland 5%-10%.[6,8,9] The incidence is slightly more in females than in males (2:1 ratio) and the aetiology is not known for certain. Pleomorphic adenomas can arise at any age but are somewhat more common between the ages of 40 and 60 years.[5-10]

This neoplasm characterised by proliferation of parenchymatous glandular cells along with myoepithelial components, having a malignant potentiality.

Malignancy it's extremely rare of the salivary glands occurs 2-5% of all neoplasms head and neck.[1,2,9,10] These tumors do not invade into periosteum.

Diagnosis of neoplastic lesions of the major salivary glands can be challenging, and therefore appropriate imaging is essential for making a provisional and correct diagnosis.[8,10] Differentiation between benign and malignant lesions or inflammatory disease may be possible with minimally invasive and cost effective procedures like FNAB.[10] The differential diagnosis includes adenoid cystic carcinoma, monomorphic adenoma, facial nerve schwannomas and polymorphous low grade adenocarcinoma, non specific neoplasms to salivary glands also. The cytologic features of salivary gland tumors, as well as some of the more commonly encountered problems and pitfalls associated with FNA interpretations are presented.[2,5-7]

On the other side, Gudmundsson JK, Ajan A, Abtahi J in their clinicopathological study of 114 patients reports that the accuracy of FNAC depends on the pathologist's experience and precision.[10]

In our case the CT scan revealed a large well defined mass in the left submandibular space with no evidence of fixation to the mandible or the floor of oral cavity. She had a mobile, soft mass with gradually enlarging neglected for more than 22 years. Pain due to benign tumor is reported in 2%–11% cases.

The cevico-submaxillary approach due surgical excision (wide

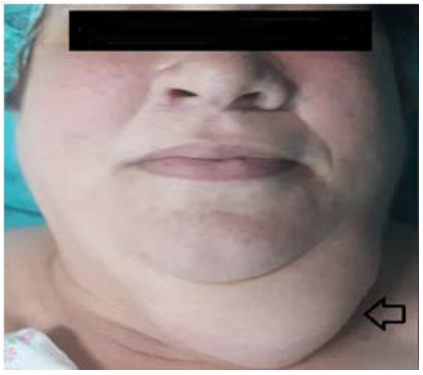


Fig. 1 Extraoral frontal view - lobulated mandibular mass (Left black arrow)

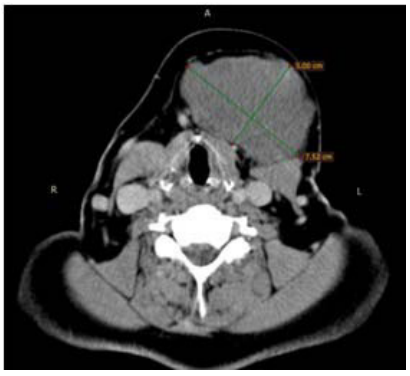


Fig. 2 Axial view Computed Tomograph: A well defined soft tissue mass in the left submandibular area.

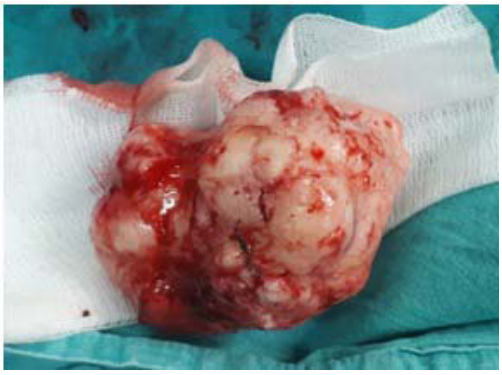
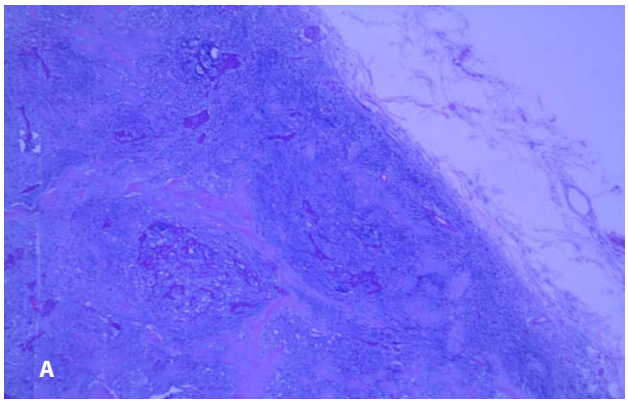
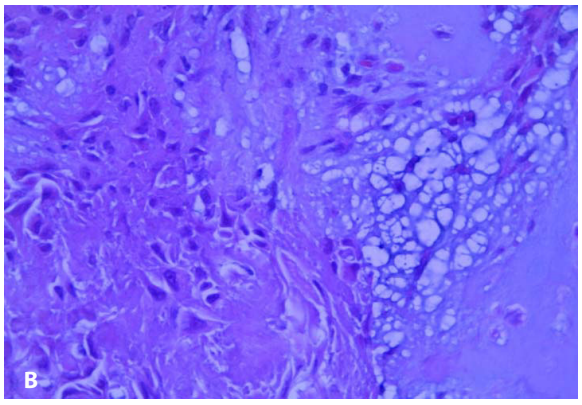


Fig. 3 Gross specimen - A well - delineated solid lesion





Figs. 4A and 4B: A typical biphasic neoplasia with a myxochondroid stromal and epithelial component without significant atypie or mitotic activity (H and E stain, 10X and 40X)

local tumour resection with safety margins particularly in present case of cheek) is the treatment of choice for PA. Prognosis is excellent and recurrence rate is low and don't vary according to age or gender. Even pleomorphic adenoma is associated with good prognosis and regular ultrasound monitoring is a prudent postoperative practice. It is noted that expression of Ki 67 predictive pathology marker is strongly associated with cell proliferation and is useful key in some cases.

Conclusion

In conclusion, the present report describes a remarkable case of PA in submandibular gland. Early diagnosis and prompt treatment is necessary to prevent the risk of recurrence rate. Continuous follow-up of patient is imperative because of the malignant transformation reported in the literature.

References

1. Ryan P. Lau, Melissa Yee-Chang, and Amy Rapkiewicz. Educational Case: Head and Neck Neoplasia Salivary Gland Tumors. Acad Pathol. 2018 Jan-Dec; 5: doi: 10.1177/2374289518770655
2. Mansueto G, Falleti J, De Cecio R, Papa F, De Rosa G. Synchronous bilateral multifocal canalicular adenoma: a case report of an unusual finding. Clin Exp Dermatol. 2009 Dec;34(8):e587-9. doi: 10.1111/j.1365-2230.2009.03258.x.
3. Somma P, Lo Muzio L, Mansueto G, Delfino M, Fabbrocini G, Mascolo M et al. Squamous cell carcinoma of the lower lip: FAS/FASL expression, lymphocyte subtypes and outcome. Int J Immunopathol Pharmacol. 2005 Jan-Mar;18(1):59-64. PubMed PMID: 15698511
4. Cantone E, Di Lullo AM, Marano L, Guadagno E, Mansueto G, Capriglione P, et al. Strategy for the treatment and follow-up of sinonasal solitary extramedullary plasmacytoma: a case series. J Med Case Rep. 2017 Aug 10;11(1):219. doi: 10.1186/s13256-017-1382-4. PubMed PMID: 28793931; PubMed Central PMCID: PMC5550939vc
5. Mounir Hmidi, Hicham Aatifi, Ali Boukhari, Mohammed Zalagh, Abdelhamid Messary. Pleomorphic adenoma of the soft palate: major tumor in a minor gland. Pan Afr Med J. 2015; 22: 281. Published online 2015 Nov 23. doi: 10.11604/pamj.2015.22.281.5227
6. Lorena Pingarrón-Martín, L. J. Arias-Gallo, G. Demaría-Martínez, M. Chamorro Pons. Remarkable Triple Pleomorphic Adenoma Affecting both Parotid and Submandibular Glands. Craniomaxillofac Trauma Reconstr. 2015 Jun; 8(2): 129–131. Published online 2014 May 29. doi: 10.1055/s-0034-1378184
7. İnan S, Aydın E, Babakurban ST, Akçay EY. Recurrent Pleomorphic Adenoma of the Submandibular Gland. Turk Arch Otorhinolaryngol. 2016 Mar;54(1):43-46. doi: 10.5152/tao.2016.1281. Epub 2016 Mar 1.
8. La Macchia R, Stefanelli S, Lenoir V, Dulguerov N, Pache JC, Becker M. Pleomorphic Adenoma Originating from Heterotopic Salivary Tissue of the Upper Neck: A Diagnostic Pitfall. Case Rep Otolaryngol. 2017;2017:5767396. doi: 10.1155/2017/5767396. Epub 2017 Nov 15.
9. 9. Endo Y, Ohashi R, Inai S, Yokoshima K, Nakamizo M, Shimizu A, et al. Carcinosarcoma ex Pleomorphic Adenoma of the Submandibular Gland in a 64-Year-Old Man: A Case Report. J Nippon Med Sch. 2018;85(1):51-55. doi: 10.1272/jnms.2018_85-8.
10. 10. Gudmundsson JK, Ajan A, Abtahi J. The accuracy of fine-needle aspiration cytology for diagnosis of parotid gland masses: a clinicopathological study of 114 patients. J Appl Oral Sci. 2016 Nov-Dec; 24(6):561-567