International Surgery Journal
Kumar MSK et al. Int Surg J. 2020 Mar;7(3):900-903
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20200842

Case Report

Uncommon occurrence of pleomorphic adenoma in submandibular salivary gland in old age males: case series

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Received: 30 November 2019
Revised: 04 February 2020
Accepted: 05 February 2020

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ABSTRACT

Pleomorphic adenoma is the most common tumor of the benign salivary gland neoplasms, the submandibular gland is the second most common site of PA after the parotid gland. Authors present 3 series case of pleomorphic adenoma in submandibular salivary gland in institution which were admitted in institution within a month interval. Fine needle aspiration cytology (FNAC) of all 3 cases proved to be benign lesion arising from submandibular salivary gland. All 3 cases underwent excision in to and the postoperative period was uneventful. DT removed on 3rd POD and discharged in POD 10. Biopsy report proved to be pleomorphic adenoma in all cases. past studies showed pleomorphic adenoma most commonly occurs in the parotid gland and its occurrence in the submandibular salivary gland is uncommon. Also, age occurrence involves 30s-50s and is more common in females. But all this case was male and occurred in older age group. Early intervention with surgical excision in toto after definite confirmation with FNAC is the treatment of choice in preventing its malignant transformation.

Keywords: Carcinoma ex pleomorphic adenoma, Pleomorphic adenoma, Submandibular salivary gland

INTRODUCTION

Pleomorphic adenoma (PA) is the most common tumor of the benign salivary gland neoplasms, Salivary gland tumors are rare and make up to 3% of head and neck tumors. 90% benign neoplasm of the major salivary gland occurs in parotid gland. PA comprises 80-90% of all benign parotid neoplasms. PA of the submandibular gland is 10% and sublingual gland is quite uncommon (0.5%) and comprises rest (<0.5%) of the group. The submandibular gland is the second most common site of PA after the parotid gland. It is also the most frequent benign tumor arising in submandibular gland.[1] PAs occur among all age groups and its incidence is 3.5/100,000. Incidence increases as age advances till around 65 years and after that incidence declines in old age.[2] The most frequent tumors of submandibular salivary gland are pleomorphic adenoma (36%), followed by adenoid cystic carcinoma (25%), mucoepidermoid carcinoma (12%), and malignant mixed tumors (10%).[3] Authors present 3 case series of a histologically proven PA involving the submandibular gland.

CASE REPORT

Case report 1

41 yr old male patient admitted with c/o swelling over the right side of neck below the jaw 1½ yrs which was insidious in onset and gradual in progression. He had no h/o pain over the swelling, no h/o LOW/LOA/fever/ trauma, pain during food intake, drooling of saliva or any other swellings. Patient was non-diabetic, not a known hypertensive, no previous hospitalization. Patient is a

known alcoholic for 10 yrs. Patient does not smoke or a tobacco chewer. No relevant family history.

Patient was well built, and his vitals were stable. no generalized lymphadenopathy, on inspection, swelling of size 4×4 cm seen in right submandibular region below the angle of mandible, single. Hemispherical, Skin over the swelling normal, No deviation of angle of mouth, No deviation of tongue. On palpation single Hemispherical swelling of size 4×4 cm noted in submandibular region extending 3 cm posterior to symphysismenti anteriorly to angle of mandible posteriorly, no warmth, no tenderness, firm in consistency, transillumination/fluctuation was negative. Mobile, on extending the neck swelling becomes more prominent, doesn't move with deglutition/ protrusion of tongue, skin over the swelling normal, no induration, margins well defined. Bidigital palpation- deep lobe of submandibular gland enlarged. Oral cavity examination- opening of stenson and Wharton ducts - normal position, dental caries were present. Both carotid arteries palpable normal in position.

His routine blood investigation was normal. USG neck shows well defined heteroechoic lesion of size 3.8×2.9 cm with increased vascularity noted in right level 2 suggestive of cervical lymphadenopathy. Fine needle aspiration cytology (FNAC) showed moderately cellular, ductal epithelial cells with myoepithelial cells in chondromyxoidstroma with few lymphocytes and macrophages s/o pleomorphic adenoma. CECT neck (Figure 1) showed well defined hypodense lesion of size 3.3×3.5 cm arising from right submandibular gland pushing right carotid space posteriorly with no contrast enhancement. Patient diagnosed as benign submandibular salivary gland neoplasm probably pleomorphic adenoma. Hence proceeded with excision biopsy removing both superficial and deep lobe.

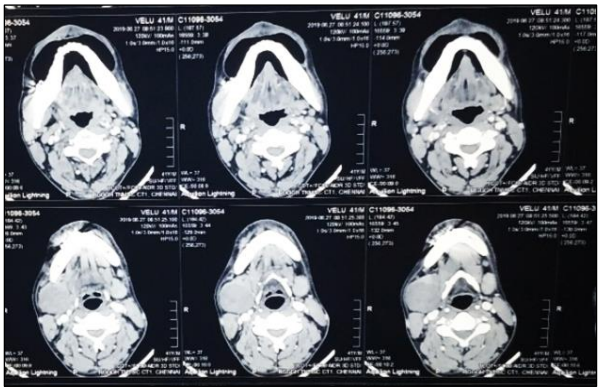

Figure 1: CT picture of case report 1.

As shown in Figure 2, 5 cm incision made 3 finger breadth below lower border of mandible, Subplatysmal flap raised, facial vessels ligated and marginal mandibular nerve lifted superiorly (Hayer Martin procedure), Superior and anterior border separated and anterior belly of digastric retracted medially to view lingual nerve and myolohyoid (Figure 4), submandibular ganglion identified and its branches to submandibular gland cut and nerve preserved, wharton duct identified and ligated with vicryl, hypoglossal nerve identified and preserved. Deep lobe visualized fully and submandibular gland excised intoto (Figure 3).

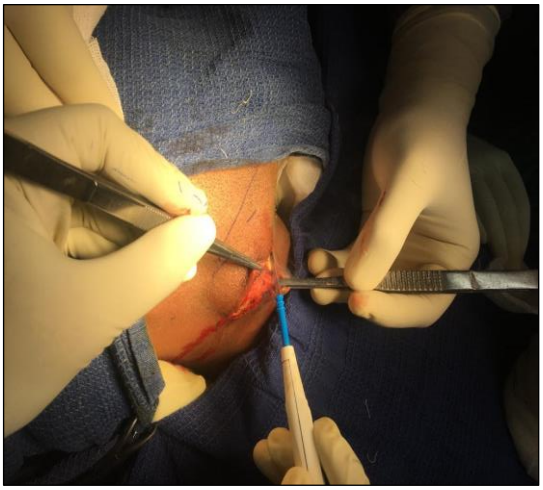

Figure 2: Incision for case report 1.

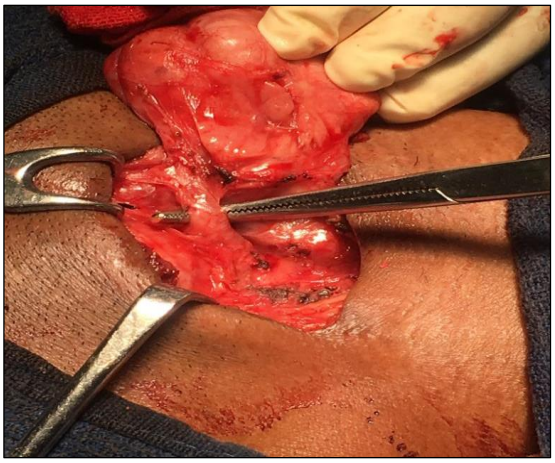

Figure 3: Dissection of case report 1.

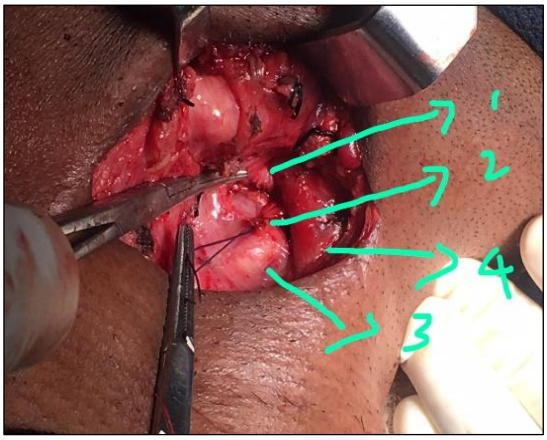

Figure 4: Intra operative picture.

Case report 2

60 yr old male patient admitted with c/o swelling over the right side of neck below the jaw 1 month which was insidious in onset and gradual in progression. He had h/o dull aching pain over the swelling for past 20 days, Pt a known not an alcoholic or tobacco chewer. Patient is a known smoker for past 26 yrs. No relevant family history.

On examination single hemispherical swelling of size 5×4 cm noted in submandibular region extending 3 cm posterior to symphysis menti anteriorly to angle of mandible posteriorly, firm in consistency. Mobile, on extending the neck swelling becomes more prominent. Bidigital palpation- deep lobe of submandibular gland enlarged. Oral cavity examination- opening of Stenson and Wharton seen - normal position, no dental caries. Carotid artery palpable normal in position.

USG neck shows well defined heteroechoic soft tissue lesion of size 2.5×3.5 cm. multiple hypoechoic nodules noted in neck of varying size. FNAC showed moderately cellular, ductal epithelial cells with matrix material and RBC's s/o benign salivary gland neoplasm probably pleomorphic adenoma. CECT neck (Figure 5) showed enlarged nodes measuring 3.3×2 cm in left side of neck s/o lymphadenitis. Patient diagnosed as benign submandibular salivary gland neoplasm probably pleomorphic adenoma. Hence proceeded with excision biopsy removing both superficial and deep lobe.

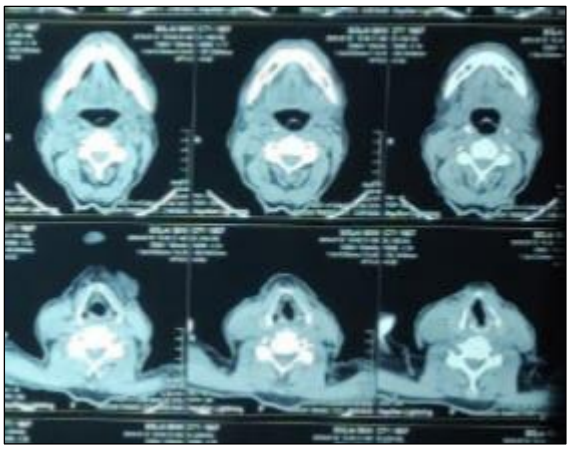

Figure 5: CT picture of case report 2.

Case report 3

55 yr old male patient admitted with c/o swelling over the left side of neck below the jaw 6 months which was insidious in onset and gradual in progression. Patient is not a smoker or alcoholic or a tobacco chewer. No relevant family history.

On examination single oval swelling of size 3×3 cm noted in submandibular region extending 4 cm posterior to symphysis menti anteriorly to angle of mandible posteriorly, firm in consistency, Mobile, on extending the neck swelling becomes more prominent, bidigital palpation- deep lobe of submandibular gland enlarged. Oral cavity examination normal. opening of stenson and Wharton seen - normal position, dental caries was present. both carotid artery palpable normal in position.

USG neck shows well defined heteroechoic lesion of size 2.1×1.5 cm lesion noted in left submandibular region suggestive of submandibular lymphadenopathy. FNAC showed moderately cellular, ductal epithelial cells with myoepithelial cells in chondromyxoidstroma with RBC s/o pleomorphic adenoma. Patient diagnosed as benign submandibular salivary gland neoplasm probably pleomorphic adenoma. Hence proceeded with excision biopsy removing both superficial and deep lobe.

DISCUSSION

Pleomorphic adenoma is usually seen in middle aged women.[4] These lesions are usually solitary, ovoid, well defined, slow growing, painless masses. The larger tumors may have pedunculated outgrowths from the main lesion and seem like multiple masses on examination.[5] Pleomorphic adenoma is an epithelial tumor comprising epithelial and myoepithelial elements intermingled with mucoid, myxoid, or chondroid tissue arranged in a variety of patterns and embedded in a mucopolysaccharide stroma.[5]

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.[4] CT scan or magnetic resonance imaging (MRI) are the gold standard radiological tools for lesion arising from both major or minor salivary glands. other investigations like ultrasound guided needle aspiration or fine needle aspiration are non-confirmatory.[1] Incision biopsy for large lesions. The recommended surgical approach is with a direct submandibular incision which provides an easy access.[1] The excision of the tumor should also be accompanied by the removal of the submandibular gland in toto. Incomplete removal of the glandular tissue paves the way for a definitive recurrence.[1]

The histological subtypes are 1. classic (mixed) 2. stroma-rich/myxoid and 3. cellular. submandibular PAs are characterized by consistent presence of an intact anatomical capsule, infrequent occurrence of pseudopodia, a remarkably infrequent occurrence lower frequency of secondary satellite tumor nodules and a comparatively lower proportion of the fragile risky myxoid subtype.[6]

The final pathologic diagnosis is always established based on findings from surgical excision. The treatment of choice for submandibular gland PA is total submandibular gland excision along with tumor.

Recurrence rate of submandibular gland tumors are less than parotid gland since entire gland is excised. Injury to the marginal mandibular nerve is the most common complication leading to temporary or permanent paralysis due to the stretching or compression of the nerve. Temporary paralysis may resolve spontaneously within a period of 3 months.[7]

Parag et al study showed, complications like neuropraxia/paresis developed in marginal mandibular branch of facial nerve in 5 (10%) patients within first 24 hours postoperatively and while it completely resolved within 3-6 month. Other complications which developed were hematoma (2%) and surgical site infection (6%).[7]

Osunde et al study showed malignant changes can occur in PA and include three distinct pathologic entities: Carcinoma arising in PA, which has been reported to occur in about 3-4%, carcinosarcoma, and benign metastasizing PA. Malignant changes were found in only 10% of the giant PA cases reviewed by Schultz-Coulon. The incidence of malignancy frequently shows a correlation between the length of the history of PA and the development of a carcinoma. It has been reported that the risk of development of malignancy is only about 1.5% up to 5 years but increases to 9.5% after more than 15 years. Hence early intervention is necessary to avoid its larger growth and preventing malignant transformation.[8]

CONCLUSION

In the past few studies that have been conducted exclusively on submandibular gland and the clinical findings in the present cases are in agreement with findings of the existing studies with PA being most common benign tumor affecting submandibular gland, occurring commonly between the 3rd and 5th decade of life and presenting as slow growing asymptomatic swelling. It is most common in females with ratio 3:1. Authors had series of case that all the pleomorphic adenoma came in male patients. 2 cases presented in old age and 1 case in middle age group and pleomorphic adenoma is common in middle age. 1 case had long duration history of swelling which itself a risk factor for malignant transformation. Both old age and male sex are risk factors for the development of carcinoma ex pleomorphic adenoma. even though m/c cause of submandibular gland swelling is sailoadenitis, authors should always include non-inflammatory lesion also in old age. Authors recommend avoiding any undue delay in management of noninflammatory lesions in the submandibular region. Preoperative FNAC proven diagnosis is highly accurate. Surgical excision is advocated as treatment of choice because of chances of malignant transformation in future. Most importantly post-operative follows up is necessary in old age and males in view of malignant transformation. If it occurs, authors have to proceed with complete surgical excision followed by radiotherapy. All 3 patients are in follow up with us & till now no new lesions noted.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Kumar MSK, Shyamsundar R, Girieasen MS. Kannan R, Nedunchezhiyan S. Uncommon occurrence of pleomorphic adenoma in submandibular salivary gland in old age males: case series. Int Surg J 2020;7:900-3.