



Pleomorphic Adenoma of minor Salivary Gland arising from Thyroglossal Duct: Uncommon Presentation

Authors

Dr Digant Patni^{1*}, Dr Shivprakash Mehta², Dr Auran Dehadaray³, Dr Maitri Kaushik⁴

¹Assistant Professor, ²Resident, ³Professor, ⁴Professor & HOD
Dept. of ENT, Bharati vidyapeeth Medical College, Pune, India

*Corresponding Author

Dr Digant Patni

Flat 203, Shreegunj Palace, Behind CHL hospital, RSS Nagar, Indore (452001) (MP), India

Abstract

Introduction: Patent thyroglossal tract commonly appears as a midline neck swelling in form of Thyroglossal cyst. Presentation in form of pleomorphic adenoma has not yet been reported. The aim of this study is to present a case of clinically appearing as thyroglossal cyst which was reported as pleomorphic adenoma of minor salivary gland.

Case Report: A 40 year old lady presented with midline neck swelling in suprahyoid region. Swelling moved with deglutination and protrusion of tongue. Intraoperatively there was a fibrous band which attached the cyst through the hyoid bone to the tongue. Complete swelling was removed with body of hyoid bone and was sent for histopathology examination (HPE).

Results: HPE of the mass revealed pleomorphic adenoma of minor salivary gland arising from thyroglossal duct. Literature search does not show any such evidence of pleomorphic adenoma arising from thyroglossal duct.

Conclusion: Midline neck swelling which clinically appears a Thyroglossal cyst may rarely be pleomorphic adenoma.

Keywords: Neck swelling, Thyroglossal cyst, Pleomorphic adenoma, midline neck swelling, salivary gland tumour.

Introduction

The majority of minor salivary gland tumours are malignant¹. Among benign tumors of the minor salivary glands Pleomorphic Adenoma (PA) is the commonest. There are nearly 450 to 750 minor salivary glands throughout the upper aero digestive tract². Aberrant salivary glands tissue may occur in variety of locations such as lymph node, lower neck, parapharyngeal space, mandible, hypopharynx, middle ear, sternoclavicular joint, pituitary gland, etc.³

Changes in these tissue may give rise to malignant transformation⁴. PA has been reported in the past in these heterotopic salivary glands^{2,4}. We present a rare case of PA of salivary gland presenting as a neck mass.

Case Report

A 40 year old female patient was referred to this tertiary hospital with painless swelling in upper neck in midline, gradually progressive from four years (Fig. 1). Physical examination revealed mass

in mid line, extending from upper border of thyroid cartilage just upto suprahyoid region.

The mass appeared non-tender, firm, mobile, non-pulsatile, non-fluctuant, moved with deglutination & protrusion of tongue. There was no bruit on auscultation. The examination of head and neck including nasopharynx, oral cavity, oropharynx, larynx, hypopharynx was normal.

Hematological investigation such as Complete blood count, Thyroid function test, Blood urea, Serum Creatinine, Blood sugar level was normal. Ultrasonography of neck & mass revealed a hypoechoic mass. Thyroid gland was present at its normal location. Fine needle aspiration biopsy of the patient was inconclusive.

The nature of swelling was different from thyroglossal cyst but as swelling was infra hyoid in position, moving with deglutination & protrusion of tongue; the probable diagnosis of thyroglossal cyst was considered. Thereafter the patient underwent Sistrunk's operation.

Intraoperatively, fibrous tract was identified to be emerging from cyst wall going posterior to the hyoid bone & attaching to the tongue base (Fig 2). Mass along with body of hyoid bone was excised. Fibrous tract going to tongue base was ligated (Fig 3). Specimen was sent for histopathological examination.

Histopathological examination revealed it to be pleomorphic adenoma of minor salivary gland (Fig.4). Patient was last followed up one year after the surgery and didn't have any recurrence on examination.

The case report was approved by Ethics Committee of Bharativedyapeeth medical college and the patient gave informed consent for use of his photographs and this case presentation.



Figure 1: Single, midline upper neck swelling



Figure 2 Fibrous band attached from mass going posterior to hyoid bone



Figure 3 Fibrous tract going to tongue base ligated & excised along with mass & body of hyoid

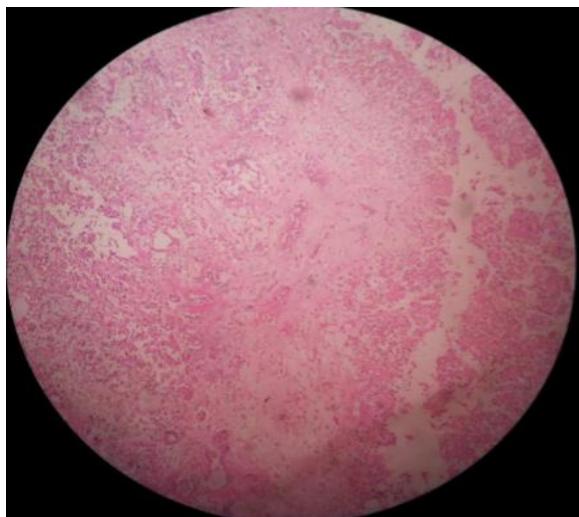


Figure 4 Epithelial cells are mostly glandular in arrangement within a myxoidstroma mostly glandular in arrangement within a myxoidstroma.

Discussion

PA, also called as benign mixed tumour, is the most common benign neoplasm of the salivary gland. They originate from an uncommitted reserve cell of intercalated duct which has potential to differentiate into epithelial & myoepithelial cells^{5,6}

PA has been reported in the extra parotid sites in the past. Palate is the most common site for pleomorphic adenoma of minor salivary gland⁷. PA has been described in maxillary sinus, tongue base, nasopharynx, retromolar area, buccal space, pterygopalatine fossa, facial skin, trachea, breast, lateral wall of nose and nasal septum⁸. PA presenting as a neck mass has been described in the past. The regions involved are submandibular⁹, upper cervical^{10,11,12}, subcutaneous plane in lower neck¹³ region. The symptoms varied from dysphagia and upper airway obstruction to asymptomatic neck mass⁹⁻¹³. In our case mass arose from thyroglossal duct presenting as a symptomatic midline neck swelling, which makes it a rare presentation.

Thus, clinically diagnostic dilemma arose in this case; as thyroglossal cyst was the topmost differential diagnosis. Though FNAC was inconclusive, excision of mass with histopathological examination confirmed the diagnosis.

Conclusion

PA of minor salivary gland arising from thyroglossal duct is rare. However, in cases presenting as a neck mass, PA should be kept in mind as a differential diagnosis due to its fairly common occurrence as a tumor of salivary gland. Although the prognosis is good, the choice of treatment in such cases is Sistrunk's operation. But, because of recurrence potential of this tumours, long term follow up is essential. Histopathological examination is the key to confirm the diagnosis and alleviate diagnostic dilemma.

References

1. Spiro RH: Salivary Neoplasms: overview of a 35 year experience with 2807 patients. *Head Neck Surgery* 1986, 8:1777-84
2. P.Peng and G.Har-El, "Anatomy and physiology", in *Essentials of Otolaryngology*, FE. Lucente and G. Har-El, Eds,pp. 3-39 Lippincott Williams &Wilkins, Philadelphia, Pa,USA,5th edition ,2004.
3. Eveson JW, Cawson RA: Tumour of the minor (oropharyngeal) salivary glands: demographic study of 336 cases.*J Oral Pathol.* 1985, 14:500-509
4. M.M Paparella and D.A.Shumrick, *Otolaryngology*, W.B. Saunders, Philadelphia, Pa,USA,3rd edition,1991
5. P.M Speight and A.W.Barrett, "Salivary gland tumours" *Oral Diseases.* 2002; 8(5): 229-40.
6. D.R. Gnepp, "Malignant mixed tumours" in *Surgical Pathology of salivary glands*, B.V. Eliss, P.L .Auclair ,and D.R.Gnepp, Eds.,pp.350-368,W.B Saunders, Philadelphia, Pa, USA,1991.
7. J.G. Batsakis, "Tumours of the major salivary glands ," in *Tumours of Head and Neck*, p.22, Williams & Wilkins, Baltimore , Md , USA,2nd edition,1978
8. Vegari et al. Pleomorphic Adenoma of the Cervical Heterotopic Salivary Gland: A

Case Report. Case Rep Otolaryngol. 2012;470652.

9. Laturiya et al. Pleomorphic Adenoma of Minor Salivary Gland Arising de novo in the Parapharyngeal Space-A Rare Case Report. Journal of Clinical and Diagnostic Research. 2016 Mar, Vol-10(3): ZD01-ZD03
10. Testa D, Staibano S, Guerra G, Mascolo M, Galera F, Iovine R, et al. Pleomorphic adenoma in ectopic salivary tissue of the neck. The Open Otorhinolaryngology Journal. 2008;2:3-5.
11. Arunkumar KV, Kumar S, Bansal V, Saxena S, Elhence P. Pleomorphic adenoma – unusual presentation of a salivary gland tumour in the neck of a child. Quintessence International. 2011;42(10):879-82.
12. Hashizume et al. Mid-cervical malignant pleomorphic adenoma presenting with upper airway obstruction. Acta Paediatr Jpn. 1990 Oct;32(5):559-62
13. Kamath B, Kamath P, Bhukebag P. Pleomorphic Adenoma in Subcutaneous Plane of the Neck: A Rare Entity. Journal of Clinical and Diagnostic Research. 2015 Sep, Vol-9(9): PD24 PD25.