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

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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 suprahoid region. Welling 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 glands2,4. 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 up to suprathyroid 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 infrahyoid in position, moving with deglutitation & 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 Bharatividyapeeth medical college and the patient gave informed consent for use of his photographs and this case presentation.
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 gland. 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 septum.6 PA presenting as a neck mass has been described in the past. The regions involved are submandibular, upper cervical, subcutaneous plane in lower neck region. The symptoms varied from dysphagia and upper airway obstruction to asymptomatic neck mass. 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


