Abstract: Objective - To report a rare presentation of basal cell adenoma in ectopic salivary gland in the neck. Case Report - A 50 year old lady with slow growing mass in upper part of anterior triangle of neck. After evaluation cytopathology was done and reported as benign salivary gland lesion. Mass was excised completely and biopsy was reported as basal cell adenoma of salivary gland origin. Conclusion - Taking into account that basal cell adenomas represent 1-2% of all salivary gland tumors, being the majority of cases in the parotid glands, the goal of this paper is to quote the rarity of the occurrence of ectopic salivary gland, and even rarer occurrence of basal cell adenoma in it. Also to sensitize clinicians to include ectopic salivary gland lesions as a differential diagnosis of lateral neck line swellings and differentiate it histopathologically from its malignant counterparts.

Keyword: Basal Cell Adenoma, Ectopic Salivary Tissue, Neck

INTRODUCTION
Basal cell adenoma is an uncommon benign salivary gland neoplasm, that appears to have unique histologic characteristics. It was included as a separate salivary gland tumor in the classification schemes by WHO in 1991. The incidence of basal cell adenoma is 1–2% of all salivary gland tumors. The most common site for a basal cell adenoma is parotid gland with 73.1% incidence in this location and seen in 20% of salivary glands especially in upper lip. The majority of occurrence is seen in patients of 60 years or older with a female predominance. It usually appears as a firm, mobile and slow growing mass. The diagnosis of this condition is solely based on histopathological studies. Treatment comprises of complete excision of the tumor. Recurrences are common especially for the membranous type. Malignant counterpart is basal cell adenocarcinoma, the chances of malignant transformation of parotid basal cell adenoma is only 0.2%.

Case Report: 50 year old female came with history of mass in left side of the upper part of neck, which was slowly and progressively growing over two years. She did not have any complaints of pain over the swelling.

INVESTIGATIONS:
1. Ultrasound study of neck showed soft cystic lesion, submandibular gland was identified separately. Impression: Query Branchial cyst/Ectopic salivary gland lesion
2. Fine Needle Aspiration Cytology was reported as benign salivary gland lesion
3. Complete blood count, Renal function test, Blood Sugar, Chest X Ray, ECG and Urine examination were within normal limits.
Per-operative findings:
Skin incision made, layers separated, mass identified deep to deep fascia, infront of Sternocleidomastoid. 1.25 cm below the ramus of mandible. By meticulous dissection entire mass was removed in toto. There was no obvious extension from submandibular gland proper. Cut section showed homogenous gray solid mass with cystic component. Specimen was send for histopathological examination.

**Fig 3** Delivery of specimen

**Fig 4** Post operative picture

Histopathology report: Multiple sections studied show an encapsulated cellular neoplasm composed of epithelial cells arranged in solid nest. The cells are basaloid showing peripheral palisading and enclosing abundant basal lamina like material. 
**Impression:** Basal Cell Adenoma of salivary gland origin.

**Fig 5** 40x magnification showing basaloid cells and eosinophilic material

Patient is on follow up for last 6 months and no evidence of recurrence has been noted.

**Discussion:** The basal cell adenoma was first reported as a separate entity by Kleinssasser and Klein in 1967. It is also referred to as tubular adenoma, trabecular adenoma, basaloid adenoma or clear cell adenoma, and it is a type of monomorphic adenoma. Batsakis is the one who first reported a case in the American literature in 1972. He suggested that the intercalated cell or reserve cell is the histogenic source of basal cell adenoma. As defined by WHO, it is a distinctive benign neoplasm composed of basaloid cells organized with a prominent basal cell layer and distinct basement membrane-like structure and no myxochondroid stromal component as seen in pleomorphic adenomas. Basal cell adenoma is a monomorphic adenoma in which basal cells predominate. It most commonly involves parotid gland (>70%), superior aspect. Peak incidence in sixth to seventh decade with female predominance. Clinically presents as a single, well defined, mobile nodule, membranous subtype tends to be multifocal. Four types of cellular patterns are seen: trabecular, solid, tubular and membranous type. Histologically, Basal Cell Adenoma is characterized by the presence of uniform and regular basaloid cells. These cells have two differentiated morphologies and are intermingled.

One group consists in small cells with little cytoplasm and intensive basaloid rounded nuclei that are usually located in the periphery of the tumoral nests or islands. The other group is formed by large cells with abundant cytoplasm and pale nuclei that are located in the centre of the tumoral nests. A basal membrane-like structure surrounds these tumoral nests, separating them from the surrounding connective tissue. The diagnosis of this entity is based on histopathology. Generally biopsy is accepted as the most accurate tool to obtain diagnosis, but some authors advice FNAC if physical access to tumor is possible. Differentiating this from similar appearing lesions is very important because some of them are malignant counter parts, like adenoid cystic carcinoma, basal cell adeno carcinoma and basaloid squamous carcinomaw.

**Review of literature:** According to Koss' Diagnostic Cytology and Its Histopathologic Bases, 5e By Leopold G. Koss, Myron R. Melamed Basal cell adenoma arise from ectopic salivary tissue at unusual sites, as for example in cervical lymph nodes and soft tissue of the neck, the breast, lacrimal glands or skin of upper and lower extremities. Mixed tumor in cervical salivary heterotopy: Authoried by Marcos Ordoñez M, Sancho Alvarez A, Morais Pérez D, Alvarez Gago T. This paper presents a case, infrequent and clinically unsuspected, of pleomorphic adenoma in the upper region of the neck in front of the sternocleidomastoid, independent of the salivary glands. With reference to an article on Pleomorphic adenoma in ectopic salivary tissue in a child written by Claros P, Turcanu D, Clarós A Jr, Clarós A, Vila Torres J published in Acta Otorrinolaringol Esp 2000 Aug-Sep;51(6):543-7, they have reported two cases of pleomorphic adenoma developing in cervical ectopic salivary tissue in children. Another case report on Ectopic salivary gland tissue in the neck. Morphology, diagnostic and histogenetic problems by French authors Auriol M, Chomette G, Bertrand JC, Devauchelle B, Delcourt A, Vaillant JM, reported 3 case of ectopic salivary gland tissue in the neck. A localized swelling was found in the upper or the middle neck along the anterior border of the sternocleidomastoid muscle. After surgical excision, the histopathological study demonstrated around a large "cystic" duct few or many lobules of salivary gland tissue in a child. They have reported one case of pleomorphic adenoma arising in cervical ectopic salivary gland tissue in a child. The embryogenesis of these heterotopies within remanants of the branchial apparatus was demonstrated in one of their cases by the association of a true branchial cleft cyst with an ectopic salivary gland. Report of a case with a review of the literature on heterotopic salivary gland tissue "Benign mixed tumour of heterotopic salivary gland tissue in upper neck" authored by Pesavento G, Ferlito A published in J Laryngol Otol. 1976 Jun;90(6):577-84. In their study the tumour was located at the angle of the right mandible, in the upper middle third of the neck, along the anterior border of the sternomastoid muscle. This observation was substantiated by a pathomorphological study. They carried out a comprehensive review of the world literature on heterotopic salivary gland tissue, together with a search of the possible pathogenesis which revealed a connection with an anomalous embryonic development within the branchial apparatus.

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Yet another study on **Heterotopic pleomorphic adenoma in the neck** by Tay HL, Howitt RJ from the Department of Otolaryngology, Ninewells Hospital, Dundee published similar details ie. Cervical heterotopic salivary tissues are rare and are predominantly found in the anterior triangle of the neck especially at the anterior border of the sternocleidomastoid muscle. Oncogenesis of heterotopic salivary tissue entrapped in an upper cervical lymph node during embryogenesis was considered as a possible etiological mechanism in this study.

**Tumours of heterotopic salivary tissue in the upper cervical region in children** by Surana R, Moloney R, Fitzgerald RJ from Children's Hospital, Dublin, Republic of Ireland From the above review of literature it is evident that ectopic salivary gland tissue is present in the anterior triangle of neck, especially in the anterior border of sternomastoid muscle, which is precisely the location of our tumour in this case.Since the mass was completely separated from all major salivary glands, and as the microscopic description was in favour of a salivary gland origin, a basal cell adenoma arising from ectopic salivary gland tissue is the most likely possibility in our case.

**CONCLUSION:**

Aim of presentation of this case is; 1. To quote the rarity of the occurrence of ectopic salivary gland, and even rarer occurrence of basal cell adenoma in it. Due to prognostic implications, differential diagnosis with basal cell adenocarcinoma, adenoid cystic carcinoma, and basaloid squamous cell carcinoma is mandatory.

To sensitize clinicians to include ectopic salivary gland lesions as a differential diagnosis of lateral neckline swellings 4. Basal cell adenoma is a specific type of monomorphic tumor of salivary glands that closely resembles basal cell lesions of the skin. Hence it is necessary to perform a complete excision of the tumor prior to the making of the final diagnosis.

**REFERENCES:**

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