

## Pleomorphic Adenoma in the Region of the Pinna : Case Report and Review of the Literature

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**A rare case of pleomorphic adenoma arising from the skin in the region of the pinna is presented.**

**Pleomorphic adenoma is the most frequent form of tumor in major and minor salivary glands. It can occasionally originate from sebaceous glands, sweat glands, or ectopic salivary glands.**

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Pleomorphic adenoma is the most common benign salivary gland neoplasm of both major and minor salivary glands<sup>1</sup>. Its occurrence in the ectopic salivary tissue and the skin is very rare. While pleomorphic adenoma is occasionally associated with cystic changes or hemorrhagic necrosis, spontaneous infarction appears to be very uncommon<sup>2</sup>. The etiology of this tumor is unknown. It arises either from myoepithelial cells or ductal reserve cells. It appears well circumscribed although may have infiltrative borders microscopically. Clinically, the tumor presents as slow growing mass and unless completely excised will recur<sup>3</sup>.

Heterotopia of salivary tissue represents developmental disorders. It is called also ectopia, which means normal salivary tissue at a site where normally not present, usually in the head and neck. The reason of its occurrence is due to misplacement of salivary gland rests cells along embryologic pathways of migration during development, or by differentiation of remnants of primitive embryologic structures. The ectopic salivary tissues may undergo same pathological processes as usually salivary gland tissue<sup>4,5</sup>.

Mixed tumors of the skin (pleomorphic adenomas) are rare benign neoplasms, which are composed of different tissue components e.g. epithelial and glandular elements and myxoid or chondroid components<sup>6</sup>.

A case of pleomorphic adenoma arising in the pinna of fifty-four years old women is presented with review of the literature. The characteristics of pleomorphic adenoma arising in areas other than the major and minor salivary glands tissue are reviewed.

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## THE CASE

Fifty-four year old Bahraini female presented with a twenty years history of a lump in the left pinna. The mass was painless and had been enlarging gradually. On physical examination, a firm non-tender subcutaneous, 4 cms mass with a pedicle arising from the antihelix of the left pinna. (Fig1). The rest of physical examination and laboratory data were not contributory to the case under discussion. Excision-biopsy of the lesion and overlying adherent skin was performed under local anesthesia. At surgery, the lesion was firm, embedded in the subcutaneous tissue and not adherent to the cartilage of the pinna. The defect was closed primarily. Post-operative period was uneventful and the patient was discharged from the hospital and kept under regular follow-up in the out-patient clinic. Follow-up at 18 months post-operatively showed no signs of recurrence. The operation site healed very well by primary intention (Fig 2). Laboratory examination of the mass removed revealed to be a brownish firm mass measuring 4x2.5x1.5 cms covered on one side by normal skin. Histological examination of the specimen showed the typical biphasic appearance due to admixture of ductular epithelium and fibromyxoid stroma interspersed in foci by cartilage. Some epithelial areas showed focal squamous metaplasia with formation of small epidermal cysts and associated non-specific granulomatous inflammation as well (Fig 3). There was no evidence of any cellular atypia and necrosis in any of the blocks studied thus supporting the nature of this so-called mixed Salivary gland tumour (syn. Pleomorphic Adenoma). The capsule was intact and there were no satellite tumor nodules detected in the surrounding tissue outside the capsule.

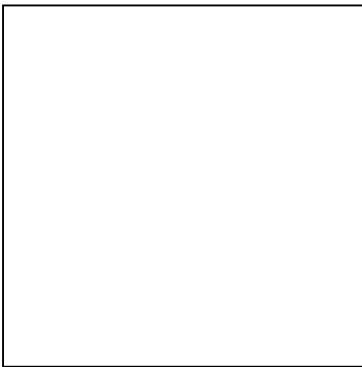


Figure 1. Pre-operative photograph of the tumour arising in the region of antihelix of the left pinna.

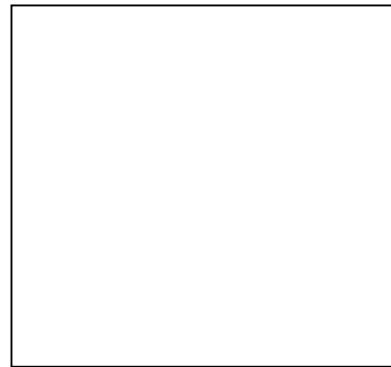


Figure 2. Post-operative photograph of the left pinna

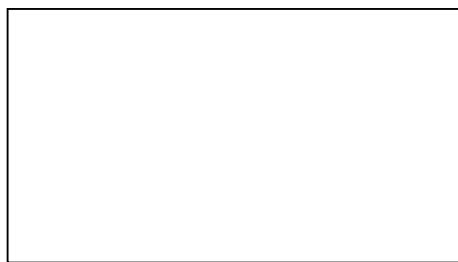


Figure 3. Micrograph of mixed tumor showing characteristic biphasic pattern of ducts and fibromyxoid stroma in between. Note, keratinized epithelial plug in a focus of squamous metaplasia (center) (Haematoxylin Eosin x 40)

## **DISCUSSION**

Pleomorphic adenomas are the most common tumors to arise in the salivary glands. They generally involve the major salivary glands, but they also can arise in the minor salivary glands and very rarely in the ectopic salivary glands and the skin.

There are four major salivary glands – two submandibular glands and two parotids. There are 300-400 minor salivary glands occurring elsewhere in the upper respiratory tract, especially in the hard palate and lateral pharyngeal wall<sup>7</sup>. Salivary gland tissue is found in the major and minor salivary glands. When an island of salivary tissue is found in a region other than the above mentioned sites, this will be referred to as ectopia or heterotopia. This may present in one of three varieties:

1. The amount of salivary tissue is large enough to cause symptoms or a mass<sup>8,9</sup>.
2. If a tumor develops within such tissue<sup>10,11</sup>.
3. When there is an associated duct and orifice discharging saliva in unusual location e.g. the skin<sup>12</sup>.

Accessory salivary glands with a duct system commonly occur anterior to the parotid gland. The ducts of the accessory salivary glands drain into the main parotid duct. These are considered variants of normal anatomy<sup>13</sup>.

Salivary glands have been described in a variety of aberrant locations including the hypophysis, cerebellopontine angle, middle ear, mastoid, auditory canal, tongue, palatine tonsil, thyroglossal duct, mandible, thyroid and parathyroid capsule, and sternoclavicular joint<sup>14</sup>. Ectopic salivary gland in the region of vulva (choristoma) and submucosa of the rectum has been reported<sup>15-17</sup>.

Pleomorphic adenomas arising in area other than the major and minor salivary glands had been reported. Cases of pleomorphic adenoma of the nasal septum have been described<sup>18,19</sup>. A comprehensive review of the world literature shows that ectopic salivary gland (ESG) tissue is rare in the region of the pinna. Only two cases in the region of the pinna have been reported in the non-English literature<sup>20</sup>.

Pleomorphic adenoma in the skin of the pinna may originate from sebaceous glands, sweat glands, or ectopic salivary glands. Various suggestions have been put forward to explain the embryological origin for ectopia or heterotopia of salivary gland tissue. The salivary glands arise by invagination of the primitive oral epithelium but only the submandibular and parotid glands migrate during their development<sup>21</sup>. This migration is limited to the area of the mandible and pre-auricular region and is unlikely to explain the presence of salivary tissue in the pinna.

Brown et al suggested that cervical lymph nodes may contain salivary inclusions, but no lymphoid tissue present in the pinna<sup>22</sup>. In branchial cleft cysts, a mucous, serous, and mixed gland had been found<sup>23,24</sup>. It has been concluded that the auricle, with the exception of the tragus and small portion of the helix at the helico-tragal sulcus derived from the second branchial arch<sup>25</sup>.

In this case, because there were no ectopic salivary glands seen on the specimen, this tumor presented here seemed to develop from the sebaceous or sweat glands of the skin of the pinna. The occurrence of pleomorphic adenoma in the skin is uncommon, and terms such as chondroid syringoma and mixed tumor have been used by some<sup>26</sup>. Chondroid syringoma was first suggested as a name for the mixed tumor of skin by Hirsch and Helwing in 1961 because the lesion is epithelial and has merely secondary changes in the stroma. The tumor develops from eccrine and apocrine glands. It usually occurs as an intradermal or subcutaneous nodule and may be attached to the overlying skin with no fixation to deeper structures<sup>27</sup>. The tumor is usually solitary and benign but malignant behavior and metastases have been reported<sup>28</sup>.

Literature reviewed found a total of 134 cases of mixed tumors of the skin. The great majority involved the skin of the face and head, including scalp, auricle, forehead, eyebrow, glabella, upper eyelid, nose, lip, chin, and neck<sup>29</sup>. Report of this case will increase the number of reported cases to 135.

The treatment of choice of a pleomorphic adenoma on the face is complete excision because incomplete resection may result in recurrence<sup>30</sup>. The patient has been regularly followed up and there is no sign of local recurrence eighteen months postoperatively.

## CONCLUSION

**A rare case of pleomorphic adenoma of the pinna is presented. Apart from its unusual location, the rare occurrence of a benign mixed tumor in the skin of the pinna (chondroid syringoma) adds to the interest of this case.**

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