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Rare case of ectopic pleomorphic adenoma on the plane of the cheek

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ABSTRACT

Pleomorphic adenoma is a benign tumor of the salivary gland with the highest incidence rate among all salivary gland tumors in the general population. Even though it's common, ectopic presentation of the same is rare. When it happens, it presents in the head and neck region. We discuss in this paper a rare case of ectopic pleomorphic adenoma on the plane of the cheek.

Keywords: Case report, Ectopic pleomorphic adenoma, pleomorphic adenoma, salivary gland tumors

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Introduction

The most common benign tumor of the salivary gland is Pleomorphic adenoma which accounts to 45-75% of all Salivary gland tumors. The most common location of presentation is the parotid gland. About 84% presents in the parotids. And about 8% in the submandibular gland and 6.5% in the minor salivary glands. The incidence is higher in Middle-aged to older women^[1] and is usually asymptomatic^[2]. It rarely presents ectopically in the head and neck region. Here, we present a rare case of ectopic pleomorphic adenoma on the cheek.

Case report:

60-year-old female presents to the Surgery department with chief complaint of a mass on the right cheek for four years duration which has been increasing in size gradually. On Physical exam the mass was 1x1 cm located in the center of the plane of the right cheek [Fig 1]. The mass

appeared firm in consistency, covered with thin skin, no discharge was observed, no punctum seen. The mass was painless. No systemic symptoms were reported. Head and neck examination were normal except the mass. Review of Systems was normal except for a history of essential hypertension. All vital signs were normal at the time of presentation. Blood cells and chemistries were ordered and came back within normal range. Sebaceous cyst and Lipoma were included in the differential diagnosis of the mass. Surgery was advised and the patient agreed. Lidocaine [2 CC] was used as a local anesthetic during the procedure. The type of incision used is transverse elliptical incision and the mass was excised. On gross examination the mass looked white, well-defined and was firm in consistency. It was sent to histopathological examination and the report confirmed the presence of a pleomorphic adenoma [Fig 2]



Figure 1: The patient presenting with mass in the right cheek

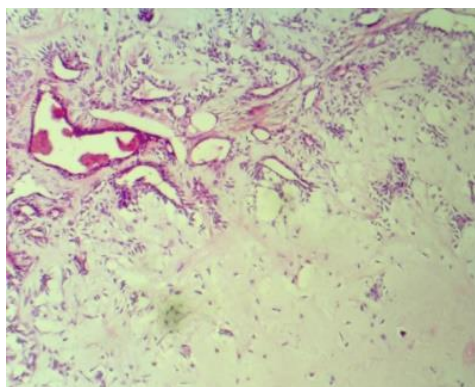


Figure 2. Histopathological report: Sections show well-circumscribed growth of both glandular and mesenchymal components composed of uniform morphology, consistent with pleomorphic adenoma [benign tumor of salivary gland]. No malignancy.



Figure 3. the 40 years old son of the patient presented with similar mass in the left cheek

Discussion

Pleomorphic adenoma, has been increasing in incidence over the past 15-20 years. It has a higher rate of occurrence in females compared to males with a ratio of 2:1. The age group of population most commonly diagnosed is between 40-60 years old [1]. Even though it's a benign tumor, pleomorphic adenoma has the potential to transform into malignant form. If identified before carcinoma develops, it is curative with resection [3]. Ectopic presentation of pleomorphic adenoma is rare and can present in the head and neck region [4]. The proposed mechanisms for this occurrence are explained by Willis which include two concepts- **Heterotopia** 1] abnormal persistence and development of vestigial structures and 2] by the dislocation of part of a definitive organ rudiment during the mass movements of developing tissue. **Heteroplasia** which is the abnormal differentiation of local tissues [5]. Heterotopia explains how glandular tissue is present ectopically but Heteroplasia explains the changes that happens in the glandular tissue that makes it grow into a tumor. Radiation exposure has been a well-known risk factor for the development of salivary gland tumors in general and also pleomorphic adenoma due to its ionizing nature [6] Another known risk factor is the simian virus [SV40], an oncogenic virus predominantly found in monkeys. In addition, there has been genetic correlation with three genes that are possibly linked to the development of this

tumor. These are the PLAG1 located on chromosome 8, HMGA2 located on chromosome 12 and the MUC1 gene. This expands the possibility that the occurrence of pleomorphic adenoma in families may be autosomal dominant in inheritance [7]. With the case that was discussed above, the son of the patient [40 years old] also presented with a similar mass on the plane of the left cheek with the same clinical features of the mass of his mother [Fig 3]. As of yet, the mass hasn't been resected and confirmation couldn't be provided regarding the histopathology of the lesion. If the son is diagnosed with ectopic pleomorphic adenoma also, this can support the idea further that even ectopic presentation of pleomorphic adenoma has a chance to run in families due to unknown genetic components yet to be studied.

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