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Original Research Paper

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ECTOPIC PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLAND PRESENTING AS CHEEK SWELLING- A CASE REPORT

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ABSTRACT Pleomorphic adenoma is the most common benign tumour of salivary glands which is known for its wide pleomorphic architecture. It accounts for 45-75% of all salivary gland neoplasm. It can involve major as well as minor salivary glands. Among minor salivary glands (5-10% of cases) the palate lip, nasal cavity, pharynx, larynx and trachea are the most common sites. Diagnosis is made with biopsy along with histopathology. Wide excision with biopsy and removal of underlying extension of tumour is the treatment of choice. We report an ectopic pleomorphic adenoma in the subcutaneous layer of the face. A 36 year old male presented with a mass of the nasolabial fold. After excision of the mass, it was revealed as a pleomorphic adenoma pathologically. An ectopic pleomorphic adenoma which was located in the subcutaneous layer of the face is very rare in medical literature.

KEYWORDS : Minor Salivary Glands; Pleomorphic Adenoma; cheek swelling; subcutaneous tissue

Introduction:

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 subcutaneous plane of the cheek.

Case presentation:

36 year old male patient came to OPD with complaints of swelling over right cheek for past 2 years which was initially smaller in size and progressed slowly to attain the present size.No other significant history was present.

On examination, swelling of size 4x3 cm in the right cheek, 4cm above the angle of mandible, spherical shape, skin over the swelling normal, buccal mucosa is normal, no deviation of angle of mouth. Swelling-well defined smooth margins, firm in consistency, not fluctuant, Not tender, able to pinch the skin over the swelling. On contracting the masseter muscle, swelling moves freely over the cheek in the subcutaneous plane. On USG, a well circumscribed hypoechogenic lesion of size 5.1x3.2cm in the subcutaneous plane of right cheek present. As followed examination, it was similar to some benign soft tissue mass such as epidermal cyst or pilomatrixoma. Blood investigations were within normal limits. The patient was then advised surgical removal of the mass.

Figure 1 (preoperative picture)

Figure 2 (preoperative picture)

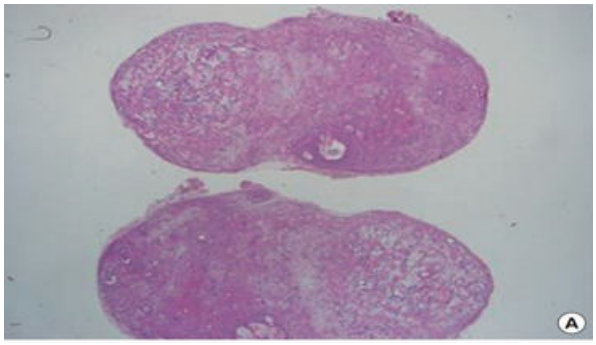
He underwent excision of the tumor (figure 3,4,5). A mass was revealed on layer of subcutaneous soft tissue. It was well defined from around normal soft tissues. Complete excision was successfully done and surgical wound was closed with primary repair. Excised mass was showed 4x3x2 cm, firm and rubbery (Fig.6). Histopathological examination revealed a pleomorphic adenoma which contains mixed epithelial and myoepithelial cells with duct-like structures (Fig.7).

Figure 3

Figure 4

Figure 5

Figure 6



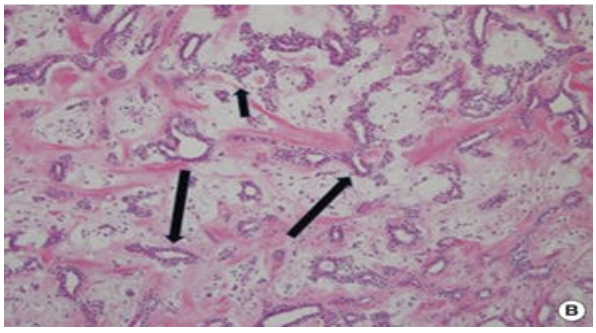


Figure 7:(A) Histopathological examination, cross-sectional inspection of a mass (H&E; magnification, ×10). (B) Pleomorphic adenoma which contains epithelial cells (short arrow) and myoepithelial cells with duct-like gland components (long arrows) (H&E; magnification, ×20)

DISCUSSION:

Pleomorphic adenomas are the most common benign salivary gland neoplasm. It accounts for 60%–70% of all parotid neoplasias, 40%–60% of all submandibular neoplasias, and 40%–70% of minor salivary gland neoplasias [1].

Majority of minor salivary glands are present in hard palate, buccal mucosa, and upper and lower lip. Other sites are floor of the mouth, inferior surface of the tongue, palatoglossal folds, and tonsillar region. Mostly it occurs between fourth and sixth decade of age, with male to female ratio varying from 1:1.4 to 1:1.7 [2]. Clinical presentation includes asymptomatic, slow growing firm mass. Small tumors typically form smooth, mobile, firm lumps but larger tumors tend to become bossellated and may attenuate the overlying skin or mucosa [3]. In our case, an ectopic pleomorphic adenoma appeared painless, soft character, it showed a clinical presentation similar to benign soft tissue tumors such as epidermal cyst and pilomatrixoma. Pleomorphic adenoma ectopically presents in the soft tissue of the neck, lymph nodes, tongue, mandible, hypophysis, mastoid bone, thyroid, parathyroid, subcutaneous layer of the nose, and skin of the external auditory canal [4]. Willis [5] proposed three main hypotheses to explain this kind of heterotopias. These were an abnormal persistence and development of vestigial structures, dislocation of portion of a deficient rudiment during mass movement and development, and abnormal differentiation of the local tissues (heteroplasia). This may happen due to metaplasia, neoplastic degeneration of ectopic salivary gland tissue or due to the implantation after surgical excision of the salivary gland tumor [6]. Pleomorphic adenoma in 5% of cases can undergo malignant transformation into carcinoma ex pleomorphic adenoma and metastasizing benign mixed tumor. Radiotherapy is not indicated due to the radioresistant behavior of the tumor. Prognosis is excellent (95%) after complete excision [7]. Thus, ectopic appearance of the pleomorphic adenoma in an unusual location can lead to a diagnostic dilemma to the surgeons and a pathological confirmation is indispensable.

Conclusion:

Pleomorphic adenoma of the cheek is a rare neoplasm. It needs to be diagnosed with a proper history and clinical examination. Wide surgical excision with a margin of surrounding tissue is the treatment of choice. Recurrence even after many years of surgical excision as well as malignant transformation should be a concern, and, therefore, long-term follow-up of these cases is advisable. Pleomorphic adenoma is a benign tumor that mainly occurs in the salivary gland. However, as in this case, a tumor that develops in the facial region needs to be differential diagnosis through a pathologic confirmation.

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