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Case Report

All Subcutaneous Swelling with Punctum are Not Sebaceous Cysts

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ABSTRACT

The most frequent benign lesion found in salivary glands is Pleomorphic-adenoma, which affects 90% of parotid glands and the remaining 10% of other salivary glands. Uncommon sites include the gingiva, cheeks, lips, and palate. Among minor salivary glands, pleomorphic adenoma occurs in the palate at a frequency of 43%, in the upper lip at 10.1%, and in the cheeks at 5.5%. It is very rare for pleomorphic-adenoma to develop on the outer aspect of the cheek. Here, we present a case of pleomorphic adenoma in the subcutaneous plane in the outer aspect of the left cheek in a 19-year-old boy. The overlying pimple mislead us to consider it as a sebaceous cyst.

Keywords: Cheek, Pleomorphic adenoma, Sebaceous cyst, Subcutaneous plane, Salivary gland

INTRODUCTION

Three percent of head and neck tumours are salivary gland tumours[1,2], and the gland that gets affected commonly is the parotid gland. Approximately 40 to 70 percent of all major and minor salivary-gland tumours are pleomorphic-adenoma, thereby making it the most frequent type of tumour.[3-6] Pleomorphic adenoma has distinct morphological and histological features and various tissues exhibit epithelial cells which are arranged in two different distinct cellular patterns - cord-like and duct-like cellular patterns, including areas of epidermoid metaplasia[3,7,8] and it also consists of a matrix that is mainly composed of myxoid, fibrous, cartilaginous, hyaline, and osseous areas.[3,7,8] The main components of pleomorphic adenoma are the capsule, myoepithelial and epithelial cells, and mesenchymal or stromal components[9], and the main function of myoepithelial cells is to contribute to the formation of pleomorphic extracellular matrix.[3] Among the minor salivary glands, the most commonly affected area is the palate (42.8% to 68.8%), followed by the upper lip (10.1%) and cheek (5.5%) in the decreasing order of occurrence.[10,11] The alveolar mucosa, the floor of the mouth, retro-molar trigone (0.7%), and throat (2.5%) are other rare sites.[10-14] The appearance of a pleomorphic adenoma is that it is slowly expanding, mobile swelling, with a firm consistency that does not cause the mucosa above to ulcerate.[10,14] Salivary gland pleomorphic adenoma that develops in the subcutaneous plane with adherence to the skin is uncommon. Most of the reported cases include the inner aspect of the cheek between the buccinator muscle and the buccal pad of fat. It is uncommon for a pleomorphic adenoma to appear directly beneath the skin without continuing into the underlying salivary glands. In this case, a 19-year-old male presented with swelling in the outer aspect of his cheek; the pimple covering the lesion deceived us into considering it as a sebaceous cyst.

CASE REPORT

A male patient aged 19 years presented to our outpatient department with a history of swelling on the outer aspect of his left cheek for the last two years. On clinical examination, a single, smooth-surfaced, spherical, movable, non-tender swelling of size 2x2 cm was noted on the outer part of the left cheek with well-defined margins. The skin on top could not be pinched. Over the swelling, several little pimples were noted that resembled the punctum of the sebaceous cyst [Figure 1].

On the left cheek ultrasonography, a well-defined multilobulated hypoechoic lesion measuring 15 x 15 x 8 mm is seen in the left buccal pad of fat. It was related to the masseter muscle laterally. It was reaching the subcutaneous plane anteriorly [Figure 2]. Fine needle aspiration cytology (FNAC) yielded inconclusive results.

Patient was posted for wide local excision of the lesion under general anaesthesia. Intraoperatively a 2 x 3 cm firm swelling was noted over the left cheek with attachment to the overlying

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Figure 1: (a) Clinical image showing swelling on the outer aspect of left side of face (oblique view); (b) Clinical image showing swelling on the outer aspect of left side of face (Side profile).

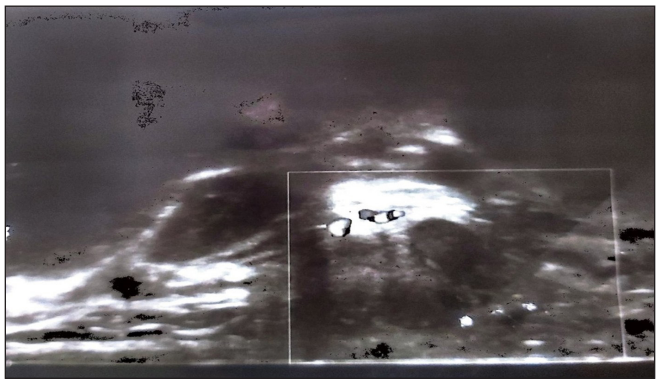

Figure 2: Ultrasonography of left cheek showing a well-defined hypoechoic lesion in the region of left buccal pad of fat.

skin. So, the overlying skin was removed along with the tumour to prevent recurrence [Figures 3 and 4].

Histopathological report was suggestive of tumour tissue comprising of epithelial, myoepithelial and stromal components. The epithelial and myoepithelial components were arranged in sheets, and at areas form cysts and tubules in myxoid stroma. The stroma was chondroid, and at areas showed myxoid change. A thin fibrous capsule surrounds the tumour [Figure 5].

The post-operative period was uneventful, with a cosmetically acceptable scar [Figure 6].

DISCUSSION

Twenty-two percent of salivary gland neoplasms arise from minor salivary glands, out of which 82% are malignant and the remaining 18% are benign lesions.[14,15] Among the minor salivary glands, pleomorphic adenoma most commonly occurs in the palate. Other sites of occurrence are, in decreasing order, the tongue, tonsils, pharynx, lips, buccal mucosa, the floor of the mouth, and retromolar area.[15,16] This condition is more common among adults aged between the

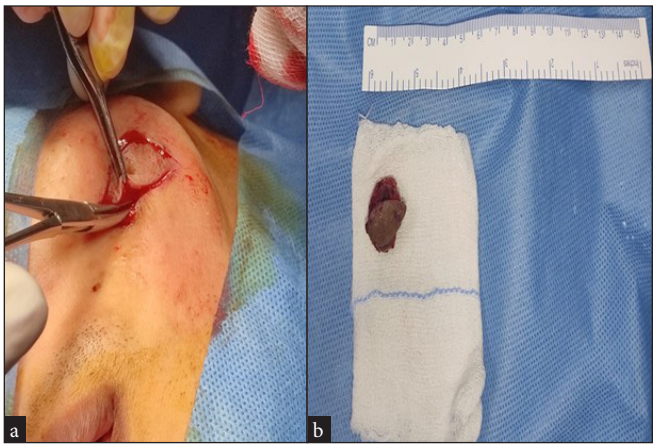

Figure 3: (a) Intraoperative image of wide local exicision of the mass; (b) Specimen photograph.

Figure 4: Incision sutured in two layers using 3-0 ethilon and 4-0 catgut.

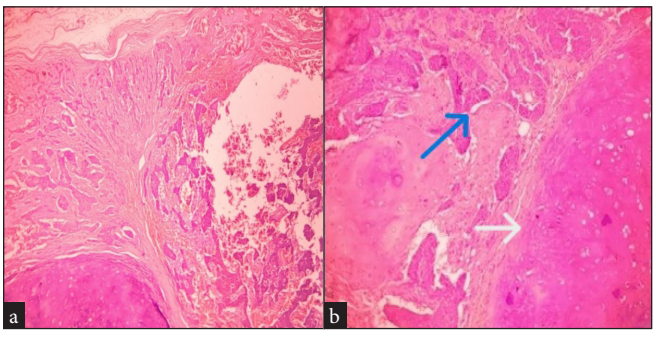

Figure 5: 10x and 40x hematoxylin and eosin stained microscopic examination of the specimen showing epithelial, myoepithelial, and stromal components. (a) Blue arrow: mesenchymal chondroid matrix (b) White arrow: epithelial cells surrounded by mesenchymal cells.

fourth and sixth decades of life.[3,14] However, the aetiology of pleomorphic adenoma is unknown. There are evidence

Figure 6: Post-operative image after 1 week. Sutures removed.

and studies which suggest that the clonal chromosome abnormalities, including 8q12 and 12q15 aberrations being associated with the aetiology of pleomorphic adenoma, which is said to be of epithelial origin.[17,18]

In most cases, the patient presents to the outpatient department with painless, gradually progressive, mobile, lobulated submucosal swelling.[10,19] Depending on the presence of degeneration, which may be myxoid or cystic, and the formation of osteoid or chondroid tissue, the consistency of swelling changes from soft and fluctuant to rubbery and firm.[19] Clinical features such as a rapid increase in the size of the swelling, discomfort, involvement of skin and other deeper layers, and lymphadenopathy indicate malignant transformation.[17] In our instance, the patient sought treatment because of a cosmetic defect that was noticeable.

Pleomorphic adenomas are complex tumors embedded in a mucopolysaccharide matrix consisting of epithelial and myoepithelial elements.[15,17] They often appear to be surrounded by a pseudo capsule that is formed by fibrosis and compression of the surrounding parenchyma.[15,17]

When combined with the ability to evaluate the degree of bone involvement, radiological tests like Computed Tomography, Magnetic Resonance Imaging, and ultrasonography are also useful in assessing the size as well as the extent of the lesion.[14,19] FNAC can also be used as a diagnostic modality. Incisional biopsy is contraindicated in a suspicious case of pleomorphic adenoma due to the increased risk of local recurrence.[19,20]

There are three distinct histopathological subtypes of pleomorphic- adenomatosis: these three types are called myxoid (80% Stromal component), cellular (predominance of myoepithelial cells), and mixed (also known as classic type).[16]

In our instance, we concluded pleomorphic adenoma through histopathological examination. For pleomorphic adenoma of the cheek, wide surgical excision with a sufficient margin of surrounding normal tissue is the preferred course of treatment.[3,15-17,19,21] Since these tumours often contain a microscopic pseudopod-like extensions that invades through the capsule into the surrounding tissues, there is increased risk of local recurrence as a result of inadequate or incomplete resection, capsular rupture, or tumour leakage during surgical excision.[3,15,17,19,22] Patients with minor salivary-gland tumours should be followed up regularly because these tumours have the potential for malignant transformation and have a higher likelihood of local recurrence.[3,16,17]

CONCLUSION

Pleomorphic adenoma of the outer aspect of the cheek in the subcutaneous plane lateral to the muscle with adherence to the skin is uncommon, and few cases have been reported. Pleomorphic adenoma should be considered as a differential diagnosis for any patient who complains of subcutaneous swelling over the cheek.

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