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CASE REPORT Open Access

Pleomorphic adenoma of the cheek: a case presentation

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Abstract

Background: Pleomorphic adenoma, also known as a benign mixed tumor, is the most common salivary gland tumor. The parotid gland accounts for 90% of the total, with the minor salivary glands accounting for 10%. Buccal minor salivary glands pleomorphic adenoma is extremely rare. It manifests as a painless, firm, slow-growing mass in most cases.

Case presentation: A pleomorphic adenoma in the buccal minor salivary glands was discovered in an adult 43-year-old female patient treated with wide surgical resection. A literature review of the PA of the cheek is stated.

Conclusion: In the differential diagnosis of cheek masses, pleomorphic adenoma should be scrutinized. The remedy of choice is wide local excision with at least a 5-year follow-up.

Keywords: Minor salivary gland tumor, Pleomorphic adenoma, Cheek, Buccal mucosa

Background

Salivary gland tumors are a broad group of tumors with a variety of clinical, histological, and immunohistochemical features [1]. These tumors are extremely rare, accounting for 3% to 5% of all neoplastic processes in the jaws [2]. They are benign neoplasms in 64.9% to 67.5% of cases [3, 4]. Minor salivary gland neoplasms, including those of the cheek mucosa, lip, and tongue, are exceptionally rare [3–5].

Pleomorphic adenoma (PA) is the most common benign salivary gland tumor, accounting for 33.2% to 68.4% of all cases [4, 5]. The parotid glands are the most affected, while the cheek, lip, and tongue mucosa are rarely affected [2, 4–6]. Females between the ages of 40 and 50 are more prone to get PA [4–7].

PA is a clinically well-defined, slow-growing, asymptomatic lesion with hard consistency and varied diameters [8, 9]. The majority of intraoral PA is solid or rubbery and is found in the submucosa. Although ulcerations are seen in certain cases, the mucosal lining stays intact [8, 10].

PA can clinically mirror other salivary gland reactive diseases and nonneoplastic proliferative processes in these intraoral locations sensitive to stress, such as the cheek mucosa, lip, and tongue.

A rare case of PA in the cheek is presented. PA's etiology, clinical and morphological characteristics, ad differential diagnoses are introduced in this context.

Case presentation

A 43-year-old female patient presented to Otorhinolaryngology Department, Kafrelsheikh University Hospital, complaining of a painless, slowly growing tumor in his right cheek 12 years ago.

A swelling of 6×2 cm in the buccal mucosa on the right side of the face was detected; it was mildly uncomfortable and freely mobile over the underlying structures and firm in consistency (Fig. 1). We could easily palpate the mass under the skin. In addition, it was mobile, nonreducible, and unattached to the underlying structures.

The color and texture of the cheek mucosa were both normal on inspection. The cranial nerves examination was revealed to be normal.

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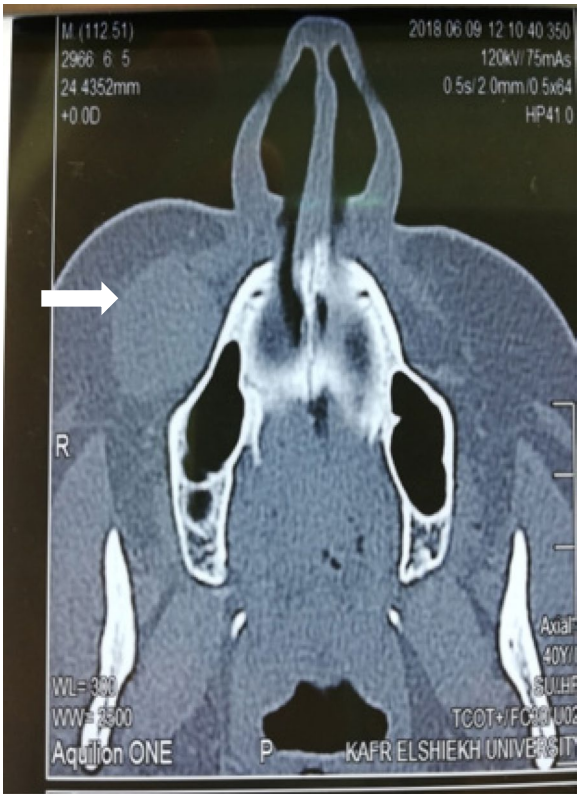

Fig. 1 CT scan nose and PNS axial section: showing well-defined lesion in the right buccal space. The white arrow points out to the lesion

The patient had a normal mouth opening. The lesion could easily be felt between the cheek mucosa and the skin with bimanual palpation. It was firm and slippery, with a slightly irregular surface. It was not fixed to the tissue around it. The lymph nodes examination was normal, with no enlargement.

A CT scan of the face revealed a well-defined mass in the right buccal space, not invading the neighboring structures (Fig. 1). The clinical assessment of the swelling has straightforwardly directed us to a provisional diagnosis of "fibrolipoma."

After the patient approved appropriate consent for the surgery, we planned it with the knowledge that the lesion was mostly benign and quite superficial (Fig. 2). Ethical Committee approval of the institute was waived.

The surgical operation was under general anesthesia under aseptic conditions for wide local excision of the mass and then a sub-labial approach closure of neighboring mucosa.

The tumor was simply removed. The mass instantly burst out with no attachment to neighboring structures once the mucosa and muscle layer were dissected. The

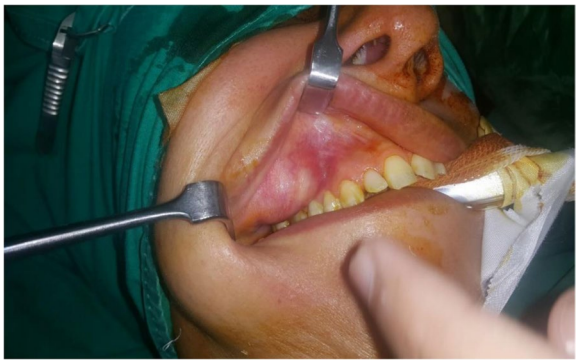

Fig. 2 Preoperative view showing a mass in the right cheek

removed mass measured 6 × 2.3 × 1.5 cm and was firm and rubbery in texture (Fig. 3).

The surgical wound was closed in layers with 3/0 vicryl absorbable sutures after the mass removal (Fig. 4). Histopathological examination was done for the specimen.

The histopathological report revealed the presence of glandular epithelium and mesenchymal tissue and changes like mucoid material accumulation, hyalinization foci, and an epithelial component made up of myoepithelial cells arranged in cords and nests, in accordance with pleomorphic adenoma characteristics (Fig. 5). Postoperative follow-up for 5 years revealed no recurrence.

Discussion

Pleomorphic adenoma (PA) is considered the most common benign salivary gland tumor [11]. Willis [12] created the name Pleomorphic adenoma. The World Health Organization (WHO) categorized PA in 1972 as "a circumscribed tumor characterized by clearly visible epithelial tissue interspersed with the tissue of mucoid, myxoid, or chondroid appearance" [13]. It strikes during the third and sixth decades of life, with a minor proclivity for

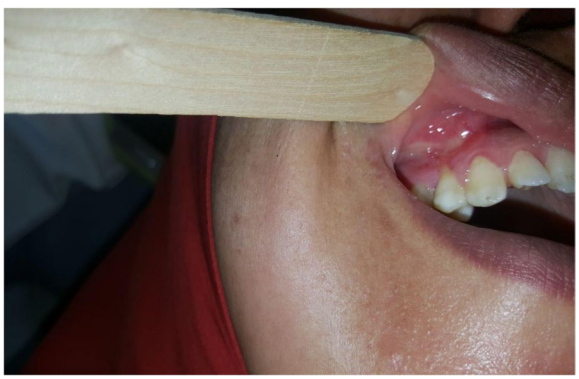

Fig. 3 Postoperative view showing healing of the wound

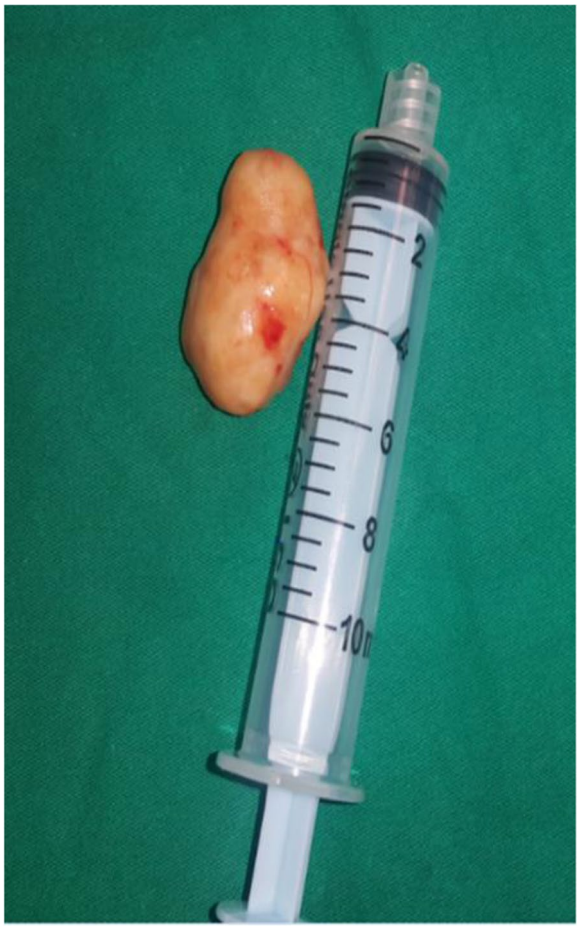

Fig. 4 Excised surgical specimen 6 × 2.3 × 1.5 cm lobular mass

females with a female-to-male ratio of 2:1 [14]. The term "mixed tumor" is deceptive [12], despite the fact that it is made up of mesenchymal and epithelial cells [15]. The parotid gland is frequently affected, particularly the lower pole of the superficial lobe. The minor salivary glands of the mouth are involved in 8% of PAs. Sixty to 65% of the salivary glands in the mouth [12] and 5.5% in the minor salivary glands of the cheek [16] are involved.

The clinical features of the lesion are consistent with those described in the literature, including an asymptomatic, slow-growing nodular lesion of firm consistency with normal intact mucosa [8–10]. Based on the characteristics of the mass as being superficial and easily accessible by excisional biopsy, we did not recommend Fine Needle Aspiration Cytology (FNAC) of the lesion preoperatively. However, FNAC is still a precise and helpful tool for distinguishing benign from malignant tumors and establishing the ideal treatment strategy.

The histopathological report of the case described the interaction of epithelial components clustered in duct-like structures with myoepithelial cells in the stroma with various patterns such as mucoid, myxoid, cartilaginous, or hyaline. These findings coincide with PA's microscopic features [8, 15].

The tumor under inquiry has a wide range of histological features that reflect the polymorphic pattern seen in salivary gland neoplasms. The histology of these malignancies, which is linked to the type of cell, handles a wide range of histomorphology [16, 17]. PA's histogenesis is linked to the intercalated duct's reserve cells. The ability of reserve cells to develop into multiple subtypes points out the morphological variability seen in salivary gland neoplasms, which can even differ within the same tumor [17].

Soft-tissue neoplasms, like neurilemoma [8, 10] neurofibroma, lipoma [8, 14] neoplasms of minor salivary glands, may also be considered for the differential diagnosis [18], in addition to inflammatory and reactive lesions [8–10].

Finally, while developing diagnostic hypotheses is critical, the significance of making definitive scrutiny is emphasized. Whereas the comprehensive look at a lesion can imply a variety of theories, the anatomopathological investigation is the only way to figure out the definitive findings. The recommended treatment for PA is surgical excision with preservation of the capsule

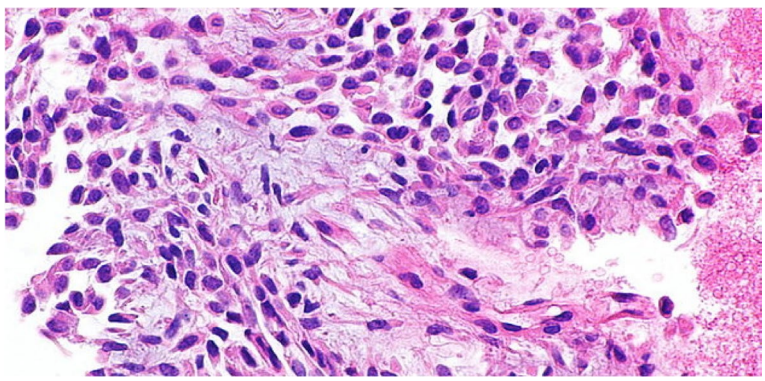

Fig. 5 Histopathology image of pleomorphic adenoma

and a perimeter of normal tissue around it [8–10, 14, 15, 19–21]. PA should be diagnosed as soon as feasible because, despite its rarity [19, 22, 23], later malignant change to carcinoma ex pleomorphic adenoma might occur [7, 11, 21–23]. Consequently, in our study, we followed up the case for 5 years with no recurrence.

Conclusion

Pleomorphic adenoma of the minor salivary gland, particularly when involving the cheek, is a rare lesion that can be difficult to detect, even for the most experienced surgeons. In the differential diagnosis of cheek swellings, it should be considered.

The recommended treatment is the surgical removal of the tumor with adequate safety margins. As a result, early detection and treatment, as well as regular follow-up for at least 5 years, are critical.

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Authors' contributions
A.S.A: data collection, writing, reference collection, editing the final draft. S. E: data collection, revision. H.E, A.A.E: final revision. M.A: review writing revision. The authors read and approved the final manuscript.

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Declarations

Ethics approval and consent to participate
Written Formal consent was signed by the participant for sharing in this research. Ethical Committee approval of Kafrelsheikh University, Faculty of Medicine, was waived. The use of any animal or human data or tissue "Not applicable".

Consent for publication
Written formal consent was signed by the participant for publication and accompanying images.

Competing interests
The authors declare that they have no competing interests.

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