

Pleomorphic Adenoma of the Inner Side of the Cheek

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DOI: [10.36347/sjmcr.2021.v09i05.004](https://doi.org/10.36347/sjmcr.2021.v09i05.004)

| Received: 17.03.2021 | Accepted: 25.04.2021 | Published: 07.05.2021

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Abstract

Case Report

The pleomorphic adenoma or mixed tumor is a heterogeneous benign tumor of salivary glands. The parotid gland is its principal localisation, it is rare in accessory salivary glands, with a preferential localisation on the oral mucosa (palate, cheeks and lips). We present a rare case of pleomorphic adenoma of the inner side of the left cheek. This is a 34-year-old patient who presents a mass on the inner left cheek that has been developing for 2 years, painless, without signs of inflammation or fistulization at the level of the oral mucosa and of the opposite skin, examination of the parotid region and peripheral lymph node areas was normal. The patient underwent an ultrasonography (USG) which revealed the presence of a left cheek submucosal mass with a polylobed tissue appearance. A surgical resection was performed and the pathological examination was in favor of pleomorphic adenoma.

Keywords: pleomorphic adenoma, salivary glands, oral mucosa, ultrasonography (USG).

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INTRODUCTION

Salivary gland neoplasms have a rate of 3e4% among all head and neck tumors. The most prevalent benign salivary gland tumor is Pleomorphic adenoma (PA) or (mixed tumor) which constitutes about 40%e70% of all salivary gland neoplasms [1]. Commonly, PA is found in the major salivary glands, besides that, it can appear in the minor salivary glands (MSG)s and extra-salivary tissue [2]. Cheek AP represent 5,5% of intraoral AP [3].

We present a rare case of pleomorphic adenoma of the inner side of left cheek.

OBSERVATION

This is a 34-year-old patient with no notable pathological history, who presents a mass on the inner face of the left cheek that has been developing for 2 years ,painless, without any notion of superinfection or fistulization, clinical examination objectified the presence of a left cheek mass ,about 4 cm long axis, with firm consistency, bumpy contours, painless, mobile in relation to the two planes, without signs of inflammation at the level of the oral mucosa and the opposite skin, examination of the parotid region and peripheral lymph node areas was normal.

The patient underwent an ultrasonography (USG) which revealed the presence of a left cheek submucosal mass with a polylobed tissue appearance.

Surgical excision of the mucosa under local anesthesia was performed.

Pathological examination of the surgical specimen showed a well-limited greyish-white tumor proliferation, polylobed on the surface and double contingent; epithelial and mesenchymal; epithelial proliferation shows epithelial and myoepithelial cells arranged in clumps, nests and tubuliform structures the mesenchymal component is enlarged chondroid and ossified in places.

The ultrasound performed two years after does not show any recurrence of the tumor.



Fig-1: Surgical excision of a pleomorphic adenoma of the inner side of the left cheek



Fig-2: Jugale pleomorphic adenoma after surgery measuring 5 cm

DISCUSSION

Pleomorphic adenoma is a heterogeneous benign tumor and the most common of the salivary glands, long called mixed tumor because of its double epithelial and mesenchymal component [4-6]. It is the most common tumor of the parotid gland (65%). Its extra-parotid localizations are rare.

They are distinguished from the parotid by their clinical, histological and therapeutic characteristics. They are distributed between the submandibular gland and the Minor salivary glands [5, 7]. Among the intraoral PA, the most frequent sites are palate (42.8e68.8%), followed by the upper lip with 10.1%, cheek with 5.5%, the throat with 2.5%, retromolar region with 0.7%. PAs may exist on the floor of the mouth and the alveolar mucosa, also, Stenson's duct, accessory parotid tissue, and parapharyngeal space are known as other ectopic presentations of PA [3].

PAs are generally asymptomatic, slow-growing neoplasms, but in a few cases, particularly in the palate, rapid growth have been noted [8]. PAs have slight differences according to sites. MSGTs are commonly fixed pathologies that are smaller than parotid tumors while neoplasms of the parotid usually are mobile, solid and different in size (2e6 cm), in our patient the size was 5 cm.

Pleomorphic cheek adenoma is a lesion that appears as a well-limited, firm nodule and painless on palpation, pushing back the mucosa without ulcerating it [3, 9] (Case of our patient).

The diagnosis of PA with fine needle aspiration (FNA) is preferred routinely in adults. If adequate samples are received and the findings interpreted correctly, FNA will be an ideal, fairly accurate procedure for the diagnosis that will help design the treatment [10].

For the diagnosis of soft tissue lesions like salivary gland tumors, several scanning techniques like Computed tomography (CT), Magnetic Resonance Imaging (MRI) or USG can be used [11]. Imaging provides significant data about the location, dimension, and expansion of the mass [12]. USG is a non-ionizing, rapid, non-invasive, widely available, cost-effective diagnostic method. The common USG appearance of PA is a well-defined margin with posterior acoustic enhancement, a regular, rarely lobulated shape, homogeneous echo patterns, and lack of vascularization [2]. USG is a useable for differentiating solid pathologies from cystic lesions. However, its use in preoperative diagnosis and planning is limited. USG can be only performed for buccal and parapharyngeal space lesions among MSGTs [12]. The presentation and treatment of PA in superficial tissues is illustrated along with the application of USG in this case. The use of USG allowed us to clearly define the margins of the mass and rule out extension of the tumor into the adjacent tissues. CT scan is the best to assess the adjacent bone to the lesion.

In MRI, PA has an intermediate or low signal intensity on T1-weighted images and a more changeable T2-weighted sequence. Myxoid and cellular areas of PA affect the pattern of enhancement on MRI [2]. Although MRI provides excellent soft-tissue contrast resolution without ionizing radiation, high costs, longer scan time, not suitable for every patient are the disadvantages of the method [13].

Macroscopically, the tumor is nodular, well circumscribed or even encapsulated by a matrix conjunctiva, it is usually gray whitish in color, translucent in some places when cut [14, 15]. Histologically PAs show morphological diversity and complexity. For diagnosis of PA, epithelial and mesenchymal tissues are necessary [12]. Epithelial cells bring out ductal structures and mesenchymal components produce to myxoid, hyaline, cartilaginous, and osseous change [2]. Myoepithelial cells are responsible for production of pleomorphic extracellular matrix. PAs have three histologic subtypes [4]: cellular (myoepithelial cells predominating) [16]; myxoid (80% stroma, the stroma rich); and [5] mixed (classic) [17]. In the oral cavity, this tumor has for peculiarity of not being encapsulated and contact tumor cells with fat cells or muscle should not be required for a carcinoma infiltrating. [6.18] complete surgical excision with resection of the covering mucosa and coagulation of the tumor bed provide the definitive diagnosis and allow treatment of this remarkable tumor of the salivary glands. This complete excision is made difficult, when it comes to large tumors, by absence of the capsule and the infiltration of the accessory salivary glands between the muscle fibers and adipose panniculus. It may require the use of reconstruction flaps in case of significant mucosal excision [5, 19].

Mucosa having a nodular appearance. Its prognosis is generally good, but remains marked by a high risk of recurrence after surgery and of carcinomatous degeneration requiring an early surgical management and regular monitoring [20].

CONCLUSION

PA of the cheek is uncommon tumor, it should be included in the differential diagnosis of cheek lesions in young and adult patients. The possibility of late recurrences and malignant transformation should be regarded. So that long-term follow-up is essential [13].

Declaration of Competing Interest

The authors declare that they have no conflict of interest.

REFERENCES

- Lopes MA, Kowalski LP, Cunha Santos G, de Almeida OP. A clinicopathologic study of 196 intraoral minor salivary gland tumours. *J Oral Pathol Med.* 1999;28:264-7.
- Lingam RK, Daghir AA, Nigar E, Abbas SA, Kumar M. Pleomorphic adenoma (benign mixed tumour) of the salivary glands: its diverse clinical, radiological, and histopathological presentation. *Br J Oral Maxillofac Surg.* 2011;49:14-20.
- Dalati T, Hussein MR. Juvenile pleomorphic adenoma of the cheek: a case report and review of literature. *Diagn Pathol.* 2009;4:32.
- Mortuaire G, Bounafos G, Wacrenier A, Darras JA, Chevalier D, Desaulty A. Adénome pléomorphe endonasal. *J Fr Otorhinolaryngol.* 2002; 4:180-4.
- Nouri H, Raji A, Elhattab Y, Rochdi Y, Ait M'barek B. Adénome pléomorphe de la voûte palatine : à propos d'un cas. *Rev d'Otorhinolaryngologie et de chirurgie cervico-faciale.* 2006;302:22-23.
- Oueslati Z, Naji R. Adénome pléomorphe de la glande submandibulaire. *J Tun ORL.* 2003;11:19-22.
- Lou SM, Rich AM, De Silva RK, Ferguson MM. Pleomorphic adenoma of a molar salivary gland. *Oral Oncology Extra.* 2006 Apr 1;42(4):170-2.
- Lotufo MA, Júnior CA, de Mattos JP, França CM. Pleomorphic adenoma of the upper lip in a child. *Journal of oral science.* 2008;50(2):225-8.
- Ayoub OM, Bhatia K, Mal RK. Pleomorphic adenoma of the parotid gland: is long term follow-up needed? *Auris Nasus Larynx.* 2002; 29:283-5.
- Alshehri AA, Al-Qarni SN, Hudise JY, Sumaily IA. Juvenile buccal pleomorphic adenoma: a rare case report. *J Otolaryngol-Head N.* 2017;3:431.
- Gupta MK, Bailoor DN, Mhaske S, Raghuvanshi V, Nahar S, Raju Ragavendra T. Juvenile Pleomorphic Adenoma of the Cheek: a rare case with review of literature. *IOSR J Dent Med Sci.* 2013;6:45-8.
- Noghreyan A, Gatot A, Maor E, Fliss DM. Palatal pleomorphic adenoma in a child. *J Laryngol Otol.* 1995;109:343e5.
- [13] Dombrowski ND, Wolter NE, Irace AL, Cunningham MJ, Vargas SO, Perez-Atayde AR, et al. Pleomorphic adenoma of the head and neck in children: presentation and management. *Laryngoscope* 2018;129(11):2603-9.
- El Kohen A, Essakalli L, Kzadri M, Sefiani S, Andaloussi ZS. Adénome pléomorphe de la base de langue. *Revue de Stomatologie et de Chirurgie Maxillo-faciale.* 2007 Jun 1;108(3):215-7.
- Baek BJ, Bae CH, Kang EG, Cho HD. Pleomorphic adenoma of the nasal vestibule. *Otolaryngol Head Neck Surg.* 2009;141:535-6.
- Boumdini H, Lebbar K, Chaouir S. Adénome pléomorphe de l'espace parapharyngé : à propos d'un cas. *Rev Med du Maghreb.* 2003;112:5-9.
- Fu H, Wang J, Wang L, Zhang Z, He Y. Pleomorphic adenoma of the salivary glands in children and adolescents. *J Pediatr Surg.* 2012;47:715-9.
- El Kohen A, Essakalli L, Kzadri M, Sefiani S, Andaloussi ZS. Adénome pléomorphe de la base de langue. *Revue de Stomatologie et de Chirurgie Maxillo-faciale.* 2007 Jun 1;108(3):215-7.
- Verma K, Kapila K. Role of fine needle aspiration cytology in diagnosis of pleomorphic adenomas. *Cytopathology.* 2002;13:121-7.
- Hodge CW, Morris CG, Werning JW, Mendenhall WM. Role of radiotherapy for pleomorphic adenoma. *Am J Clin Oncol.* 2005;28:148e51.