



## Exeresis Of Volumous Pleomorphic Adenoma Clinical Case Report

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Submitted: 16-03-2024

Accepted: 30-03-2024

### SUMMARY

**Introduction:** pleomorphic adenoma is the most common benign tumor of glandular origin on the face (oral cavity). It represents approximately 70 to 90% of tumors of the major salivary glands, more rarely in the submandibular gland. **Objective:** to report a clinical case of pleomorphic adenoma located in the left submandibular gland, treated through surgical excision. **Case Report:** this article describes a relatively unusual case in a female patient, 39 years old, with slow growth (6 years). Patient without comorbidities, smoker, without pain, reported difficulty swallowing, with no previous history of trauma or infection. The tumor was excised using the Risdon approach, and histopathological analysis diagnosed pleomorphic adenoma. **Conclusion:** The patient is in a follow-up phase until she turns five years old.

**Keywords:** Benign neoplasm; pleomorphic adenoma; surgery.

### I. INTRODUCTION

Pleomorphic adenoma, also known as benign mixed tumor, is the most common benign salivary gland tumor, accounting for approximately

70–90% of all salivary gland tumors; and may also affect the palatal or labial minor salivary glands.<sup>1,2,3,4</sup> Pleomorphic adenoma was first named by Willis.<sup>5</sup> These tumors are more commonly seen in women and have a peak incidence in the fifth and sixth decades of life. The submandibular gland is the second most commonly affected after the parotid gland.<sup>6,7</sup>

Surgical excision of the tumor is the treatment of choice for pleomorphic adenomas of the salivary glands, with high success rates and low recurrence rates. The procedure removes the tumor, leaving the surrounding environment intact.<sup>8,9,10</sup>

The histological study shows that pleomorphic adenoma is a limited and typically encapsulated tumor, presenting epithelial and myoepithelial cells with a stromal background, most of the time. There may be great microscopic variation from one tumor to another and in different areas of the same lesion. There is a diverse appearance of the epithelium, with ductal and cystic structures, as well as nests or islands of cells.<sup>3,11,12</sup>

Imaging exams play an important role in the diagnosis of salivary gland neoplasms, helping



to locate and extend lesions, as well as contributing to surgical planning.

The prognosis of the pathology is considered excellent when surgery is performed appropriately, with a cure rate of approximately 95%, with a risk of transformation into a malignant lesion in 5% of cases.<sup>13,14</sup>

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Although it is a benign tumor, there is the possibility of malignant transformation into a former pleomorphic adenoma, especially in cases with a long evolution and history of recurrence.<sup>12</sup> In view of this, the importance of the correct diagnosis of this pathology and its importance within the maxillofacial surgery and traumatology. The present study aims to discuss the literature and present a clinical case.

## II. CASE REPORT

Patient ACF, 39 years old, female, with melanoderma (figure 1), sought the oral and maxillofacial surgery and traumatology service for a specialized evaluation consultation. She reported the appearance of a nodule in the cervical region almost 6 years ago, slow growth, painless and an aesthetic issue. Smoker. Denies previous comorbidities.



Figure 1. ACF patient. 39 years old. Source: the author.

On physical examination: she was in good general condition, afebrile, flushed, hydrated, anicteric, acyanotic. The extraoral examination revealed a nodular lesion of firm consistency, asymptomatic, unilocular, measuring 7 cm in height and 4.8 cm in width, in the left submandibular cervical region. Painless palpation. During the intraoral examination, the presence of

swelling or other changes was not detected. From then on, a CT scan was requested. In the axial section, it showed the extent of the lesion, its tissue relationships, with a solid mass, with cystic degeneration, septations and acoustic reinforcement (figure 2).

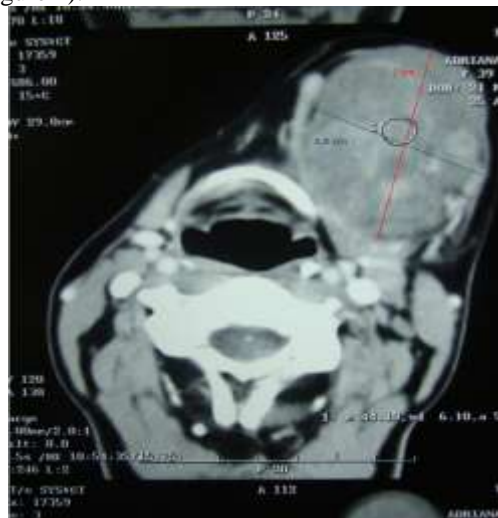


Figure 2. Computed tomography. Axial cut. Source: the author

The proposed treatment was excision of the tumor. Patient undergoing surgery at hospital level, under general anesthesia. The surgical approach was Risdon, with an incision (marking) of 1.5 to 2 cm from the lower margin of the mandible (however, the extensive volume of the tumor showed a variation in the access).<sup>15</sup> (Figure 3).



Figure 3. Left submandibular marking (Risdon Access). Source: the author.

Local vasoconstriction is performed with 1:140.000 adrenaline infiltration into the subcutaneous tissue, for local hemostasis; It should not be applied deeply to the platysma muscle, because the marginal mandibular branch of the

facial nerve will make nerve transmission non-conductible, making electrical testing impossible (if necessary).<sup>15,16</sup> (Figure 4).



Figura 4. Infiltração de adrenalina 1:100.000 (hemostasia). Fonte: o autor.

The incision was made in anatomical planes: skin, subcutaneous cellular tissue and divulsion; platysma (divulsion of the muscle by cleavage), as the most controlled method is to dissect through the platysma muscle, from one end of the skin incision with the tips of a hemostat or Metzenbaum forceps. After releasing the platysma muscle beneath the superficial white layer of cervical fascia, the tips of the instrument are brought back through the platysma muscle at the other end of the incision. The submandibular salivary gland can also be visualized through the fascia, as it helps in the formation of its capsule. Dissection through the superficial layer of the deep fascia is the step that requires the most attention because of the anatomical structures with which it is associated. The facial vein and artery are usually found during access to the area of the premasseteric notch of the mandible, as well as the marginal mandibular branch of the facial nerve. (Figures 5,6, and 7 respectively).



Figure 5. Incision by planes (skin and subcutaneous cellular tissue). Source: the author.



Figure 6. Incision and divulsion (cleavage) in the platysma muscle. Source: the author.



Figure 7. Tumor. A rough, pedunculated mass, with a hard, consistent surface. Source: the author.

The masseter and medial pterygoid muscles are sutured together with reabsorbable interrupted sutures (vicril 3.0), closing all dead spaces. The superficial layer of the deep cervical fascia does not require definitive suturing. The platysma muscle was closed with a continuous resorbable suture and subcutaneous resorbable sutures followed by skin sutures, intradermal sutures, with 4.0 nylon thread and compressive dressing. After 07 (seven) days, the patient returned to have the suture removed and her postoperative period evaluated. (Figure 8 and 9 respectively).



Figure 8. Suture by planes. Source: the author.



Figura 9. Sutura intradérmica. Fonte: o autor.

The determining factor for recurrence is not the period of evolution of the lesion during which surgical treatment is performed; the tumor recurrence rate varies according to the surgical technique used. Monitoring cases of Pleomorphic Adenoma is of great value, and follow-up should be five years.<sup>12</sup> It must be certain that all of the tumor has been removed and that no tumor tissue has remained in the surgical cavity.

### III. DISCUSSION

Postoperative follow-up plays an essential role in the evaluation of surgical procedures, as well as complementary imaging exams, to evaluate recurrences and malignancy ex pleomorphic adenoma. The patient's postoperative review showed no complications or complaints, indicating a successful surgical procedure.<sup>17,18</sup>

Pleomorphic adenoma is the benign tumor with the highest incidence in salivary glands, accounting for around 70% of neoplasms in this location. Despite its benign nature, it can cause

difficulties in clinical management, particularly due to its tendency to relapse and the risk of malignant transformation, which can vary between 6 and 13% of cases. Its origin and pathogenesis still remain uncertain.<sup>19,20, 21,22,23,24</sup>

This tumor generally affects adults between the 3rd and 5th decades, but can appear at any age, with the same clinical behavior and prevalence in females, with a frequency of 60% of cases. These neoplasms show a higher prevalence in major salivary glands, more frequently in the parotid gland (67.7% to 84%) and in minor salivary glands, where the hard palate is the most predominant location.<sup>1,4,7,12,23,24</sup>

In general, clinically, Pleomorphic Adenoma of the major and minor salivary glands may have a differential diagnosis of palatal abscess, odontogenic cyst, non-odontogenic cyst, similar to a soft tissue tumor, fibroma, lipoma, neurofibroma, neurilemmoma, lymphoma or other tumors of the glands. salivary. Palatal abscess can be differentiated by identifying the source of the palatal abscess which would be a non-vital tooth in the immediate vicinity. Odontogenic and non-odontogenic cysts do not exhibit cystic nature during exploration of the mass.<sup>25,26</sup>

The submandibular gland is the second most common site of pleomorphic adenoma, approximately 5% of cases, after the parotid gland, 80-90%. It is also the most common benign tumor arising in the submandibular gland.<sup>6,7, 27,28,29,30</sup>

Ultrasonography (US)-guided fine needle aspiration cytology and core needle biopsy are safe and rapid methods for preoperative diagnosis of superficial masses.<sup>31,32</sup>

In a study of 212 patients with pleomorphic adenoma, the sensitivity and specificity of the cytological diagnosis were 92.6 and 98.4%, respectively.<sup>30,33</sup>

Computed tomography (CT) was the only imaging method used to characterize the submandibular mass in the reported case. Although CT is a widely available method, with good spatial resolution and shorter acquisition times compared to magnetic resonance imaging (MRI), it has the disadvantage of using ionizing radiation, especially relevant in children, and a limited role in tissue characterization. mole injuries.<sup>30,34,35</sup>

Surgical excision of the nodule and affected gland is the standard treatment. However, complete excision of the entire affected gland is performed mainly in pleomorphic adenoma in the submandibular gland.<sup>30,35</sup>



#### IV. CONCLUSION

Surgical excision of submandibular pleomorphic adenoma is a safe and effective treatment option, with low morbidity rates. In this case, the success of the surgery significantly altered the patient's quality of life.

#### REFERENCES

- [1]. C. Ungari, F. Paparo, W. Colangeli, G. Iannetti. Parotid glands tumours: overview of a 10-year experience with 282 patients, focusing on 231 benign epithelial neoplasms, *Eur. Rev. Med. Pharmacol. Sci.* 12 (2008) 321–325
- [2]. Ejeil AL, Moreau, N, Le Pelletier, F. A rare ectopic localization of pleomorphic adenoma. *J Stomatol Oral Maxillofac Surg.* 2019;120(4):373-374.
- [3]. M. AlKindi, S. Ramalingam, LA. Hakeem, AlSheddi, MA. Giant parotid pleomorphic adenoma with atypical histological presentation and long-term recurrence-free follow-up after surgery: a case report and review of the literature, *Case Rep. Dent.* 2020 (2020), e8828775, <https://doi.org/10.1155/2020/8828775>.
- [4]. Mattos, PR., et al. Pleomorphic Adenoma Arising FrommFrontozygomatic Region: An Ectopic Localization. *Craniomaxillofacial Research & Innovation* Volume 8: 1–5. 2023 – DOI: 10.1177/27528464231211658
- [5]. Rajendran S, Sivapathasundaram S. *Shafer's Textbook of Oral Pathology*. 6th ed., New Delhi: Elsevier; 2009. pp. 219–224.
- [6]. Alves FA., Perez, DEC, Almeida, Lopes, OPMA. Kowalski, LP. Pleomorphic adenoma of the submandibular gland: clinicopathological and immunohistochemical features of 60 cases in Brazil, *Arch. Otolaryngol. Neck Surg.* 128 (2002) 1400–1403, <https://doi.org/10.1001/archotol.128.12.1400>. [7]
- [7]. Jain, S, Hasan, S, Vyas N, Shah, N. Dalal, S. Pleomorphic adenoma of the parotid gland: Report of a Case with review of literature, *EthiopJ. Health Sci.* 25 (2015) 189–194.
- [8]. Bokhari, MR, Greene, J. Pleomorphic adenoma, in: *StatPearls*, StatPearls Publishing, Treasure Island (FL), 2022 (accessed February 15, 2023), <http://www.ncbi.nlm.nih.gov/books/NBK430829/>
- [9]. Dulguerov, P, Todic, Pusztaszeri, JM, Alotaibi, NH., Why do parotid pleomorphic adenomas Recur? A systematic review of pathological and surgical variables, *Front. Surg.* 4 (2017) (accessed February 15, 2023), <https://www.frontiersin.org/articles/10.3389/fsurg.2017.00026>.
- [10]. Grasso M., et al. Rupture of the pleomorphic adenoma of the parotid gland: what to know before, during and after surgery, *J. Clin. Med.* 10 (2021) 5368, <https://doi.org/10.3390/jcm10225368>.
- [11]. Neville, BW. et al. Salivary gland pathology. In: Neville, BW. *Patologia oral & maxilofacial*. Rio de Janeiro: Guanabara Koogan, 2009. cap. 11. p. 373-417.
- [12]. Souza RIM., et al. Pleomorphic adenoma in the submandibular gland: case report and a review of current findings. *Rev. Cir. Traumatol. Buco-Maxilo-Fac, Camaragibe, PE*, v. 13, n.2, p. 09-14, 2013.
- [13]. Lima, GF., et al. Pleomorphic adenoma of the salivary gland: an integrative review of the main literary findings. *Revista Interdisciplinar em Saúde, Cajazeiras*, 5 (2): 357-369, abr./jun. 2018.
- [14]. Alves, LP., et al. Pleomorphic adenoma of the submandibular gland. *Brazilian Journal of Health Review, Curitiba*, v. 5, n.5, p. 18907-18915, sep./oct., 2022
- [15]. Ellis, E.III; Zide, M.F. *Surgical Access to the Facial Skeleton*. 2 ed. São Paulo: Livraria Santos Editora, 2006.
- [16]. Júnior, BC., et al. Assessment of the Marginal Mandibular Nerve Function after Risdon Approach. *Rev. cir. traumatol. buco-maxilo-fac.* vol.11 no.2 Camaragibe Abr./Jun. 2011
- [17]. Murff, HJ., et al. Relationship between patient complaints and surgical complications, *Qual. Saf. Health Care* 15 (2006) 13–16, <https://doi.org/10.1136/qshc.2005.013847>.
- [18]. Birigi, M. Mweya, CN. Surgical tumour excision of pleomorphic adenoma of submandibular salivary gland: A case report and literature review. *International Journal of Surgery Case Reports* 106 (2023) 108236.
- [19]. Gonçalves, APS, Costa, JS, Sobral, APV. Malignant transformation of pleomorphic adenoma: systematic review of the literature. *Ciências Biológicas e de Saúde*



- Unit | Facipe | v. 3 | n. 2 | p. 89-104 | Novembro 2017 | periodicos.set.edu.br
- [20]. Gnepp, DR. Malignant mixed tumors of the salivary glands: a review. *Pathol. Annu., East Norwalk.*, v. 28, p. 279-328, 1993.
- [21]. Ellis, GL, Auclair, PL. Tumours of the salivary glands. In: *ATLAS of tumor pathology*. Washington, DC: Armed Forces Institute of Pathology, 1996. p.155-175. (3rd. series, fascicle 17.)
- [22]. Eveson, JW., et al. Tumours of the salivary glands: Introduction. In: BARNES, L. et al. (Ed.) *Pathology and genetics of head and neck tumours*. Lyon: IARC Press, 2005. p. 212-215.
- [23]. Martins de Sousa, GF, Leite Ribeiro, PM, Barroso, KMA. Consideration upon histopathological aspects of pleomorphic adenoma in parotid gland: study case. *Rev. Ciênc. Méd. Biol., Salvador*, v. 18, n. 3, p. 416-420, set./dez. 2019. <http://dx.doi.org/10.9771/cmbio.v18i3.34415>
- [24]. Pei, Y, Li, W. Clinical parameters predictors of malignant transformation of recurrent parotid pleomorphic adenoma. *Scientific Reports* | (2023) 13:4543 | <https://doi.org/10.1038/s41598-023-29714-6>
- [25]. Almeslet, AS. Pleomorphic Adenoma: A Systematic Review. *International Journal of Clinical Pediatric Dentistry* (2020): 10.5005/jp-journals-10005-1776
- [26]. Xia, F, Guo, F, Liu, Z. et al. Enhanced CT combined with texture analysis for differential diagnosis of pleomorphic adenoma and adenolymphoma. *BMC Med Imaging* 23, 169 (2023). <https://doi.org/10.1186/s12880-023-01129-9>
- [27]. Laskawi R, Ellis M, Arglebe C, Schott A. Surgical management of benign tumors of the submandibular gland: A follow-up study. *J Oral Maxillofac Surg* 1995;53:506-8.
- [28]. Rai S, Sodhi S, Sandhu SV. Pleomorphic adenoma of submandibular gland: An uncommon occurrence. *Natl J Maxillofac Surg* 2011;2:66-8.
- [29]. Meira, NS., et al. Pleomorphic adenoma of the submandibular gland: a case report. *Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology* Volume 134, Issue 3, September 2022. <https://doi.org/10.1016/j.oooo.2022.01.536>
- [30]. Wu, Z., et al. Surgical Treatment of a Giant Pleomorphic Adenoma of the Submandibular Gland: A Case Report. *Front. Surg.* 8:800563. doi: 10.3389/fsurg.2021.800563
- [31]. Gonçalves, AJ., et al. Needle aspiration in salivary gland tumors: specificity and sensitivity. *Rev Assoc Med Bras* 2007; 53(3): 267-71
- [32]. Faquin, WC. et al. (ed.) *The Milan system for reporting salivary gland cytopathology*. Switzerland: Springer International Publishing, 2018. 281p.
- [33]. Viguer JM, Vicandi B, Jimenez-Heffernan JA, Lopez-Ferrer P, Limeres MA. Fine needle aspiration cytology of pleomorphic adenoma. An analysis of 212 cases. *Acta Cytol.* (1997) 41:786-94. doi: 10.1159/000332705
- [34]. Som PM, Curtin HD. *Head and Neck Imaging E-book*. Amsterdam: Elsevier Health Sciences; 2011.
- [35]. Cunha-Cabral, D, Marques Gomes P, Alves Carção A, et al. Pleomorphic adenoma of the submandibular gland in children: Case report and review of the literature. *B-ENT*. 2023;19(2):134-140.