

## Conservative Surgical Approach to Pleomorphic Adenoma in Parotid Gland: Case Report

### Abordagem Cirúrgica Conservadora de Adenoma Pleomórfico em Glândula Parótida: Relato de Caso

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#### Abstract

Pleomorphic adenoma is a benign neoplasm of the salivary glands that affects most frequently females in the fourth and sixth decade of life. As it is usually an asymptomatic lesion, it is diagnosed upon routine physical examination. Treatment consists in surgical removal with partial or total involvement of the parotid gland. Conservative enucleation is the least invasive surgical option for this type of lesion. The present work aimed to report a case of pleomorphic adenoma of the parotid gland with eight years of evolution. The tumor was treated conservatively. Patient has been followed for sixth months, and no sensory alterations, fistulae, clinical signs of recurrence, nor any other alteration have been observed so far. Due to the benign nature of the lesion, an evolution with favorable prognosis is expected.

**Keywords:** Adenoma. Benign Neoplasia. Tumor

#### Resumo

*O Adenoma pleomórfico é uma neoplasia benigna de glândulas salivares, que acomete com maior frequência pessoas do sexo feminino entre a quarta e sexta década de vida. Por se tratar de uma lesão muitas vezes assintomática, geralmente é descoberta através do exame físico de rotina. O seu tratamento consiste em remoção cirúrgica podendo envolver ou não a glândula parótida de maneira parcial ou total. A enucleação conservadora é a modalidade cirúrgica menos invasiva para este tipo de lesão. O presente trabalho tem por objetivo relatar um caso de adenoma pleomórfico em glândula parótida com oito anos de evolução. O tumor foi tratado de maneira conservadora. A paciente está sendo acompanhada há 6 meses e não foram observados até momento alterações sensoriais, fistulas, indícios clínicos de recidiva ou qualquer outra alteração. Devido ao caráter benigno da lesão, espera-se evolução com prognóstico favorável.*

**Palavras-chave:** Adenoma. Neoplasia Benigna. Tumor.

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#### 1 Introduction

Salivary glands tumors (SGT) play a prominent role in human neoplasms, although uncommon, comprise an important area of oral pathology and represent one of the most interesting and challenging tumor groups in the head and neck region<sup>1,2</sup>. SGT have an annual incidence in the west from 2.5 to 3.0 cases for every 100,000 persons<sup>3-7</sup>.

Within this group, Pleomorphic Adenoma (PA), the most common benign neoplasm of the major salivary glands (GSMA) and minor ones (GSMe) stand out<sup>3,8,9</sup>. This lesion is described as a neoplasm of slow evolution, well delimited, painless, apparently encapsulated, that rarely exceeds 5 cm in its largest diameter<sup>3,5,10</sup>. The capacity of the PA to undergo recurrences and malignant transformation has been reported by literature, especially in cases where there has been incomplete surgical excision, multiple local recurrences and in tumors that have remained for long periods without diagnosis and treatment<sup>11-15</sup>.

Although the clinical and histological aspects of this neoplasm are well known, its etiology and pathogenesis

remain uncertain<sup>4</sup>. However, it is known that in more than 70% of the cases there is an association with abnormal karyotypes, involving a locus of the *PLG1* gene (*Pleomorphic adenoma gene 1*). Recently, new genes that may be involved in tumor formation have been identified<sup>16</sup>. In addition, there is evidence that exposure to UV (ultraviolet) rays increases the risk of developing it<sup>3</sup>.

The treatment of choice for this neoplasm consists of the complete surgical excision<sup>17</sup>. In the parotid gland, when the tumor is located in the superficial lobe, it may be necessary to perform the superficial parotidectomy preserving the facial nerve. On the other hand when the tumor is located in the deep lobe, total parotidectomy is recommended<sup>7,14,18,19</sup>.

The objective of this study is to report a case of pleomorphic adenoma in the parotid gland. The lesion was surgically treated conservatively and is found to be 6 months of follow-up without showing signs of recurrence.

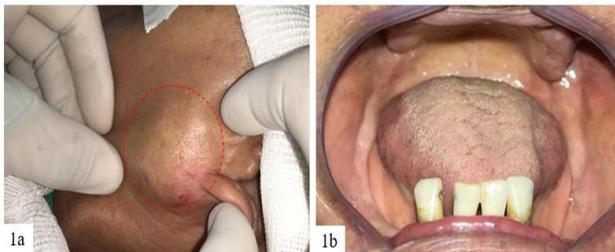
#### 2 Case Report

Patient M.L., female gender, 66 years old, parida, maid, attended the outpatient clinic of the Buccomaxillofacial

Surgery and Traumatology Service of the General Hospital of Cuiabá - MT, showing a volumetric increase on the left side face with a time of evolution of approximately 8 years and referring to mild discomfort to palpation.

In the anamnesis, she reported having controlled systemic arterial hypertension (SAH) with the use of Captopril® 25mg and Hydrochlorothiazide 25mg, both ingested once a day. Upon the extra-oral physical examination, a mobile volumetric increase was observed in the left parotid gland region of approximately 35 mm in its largest diameter, based on a sessile, delimited, of hardened and painful consistency to palpation (Figure 1a). No significant changes were observed in the intraoral evaluation (Figure 1b).

**Figure 1** - Extra and intra-oral physical examination. (1a) Delimitation of the volumetric increase in the left parotid gland region; (1b) intraoral view demonstrating integrity of the remaining mucous membranes and dental elements.

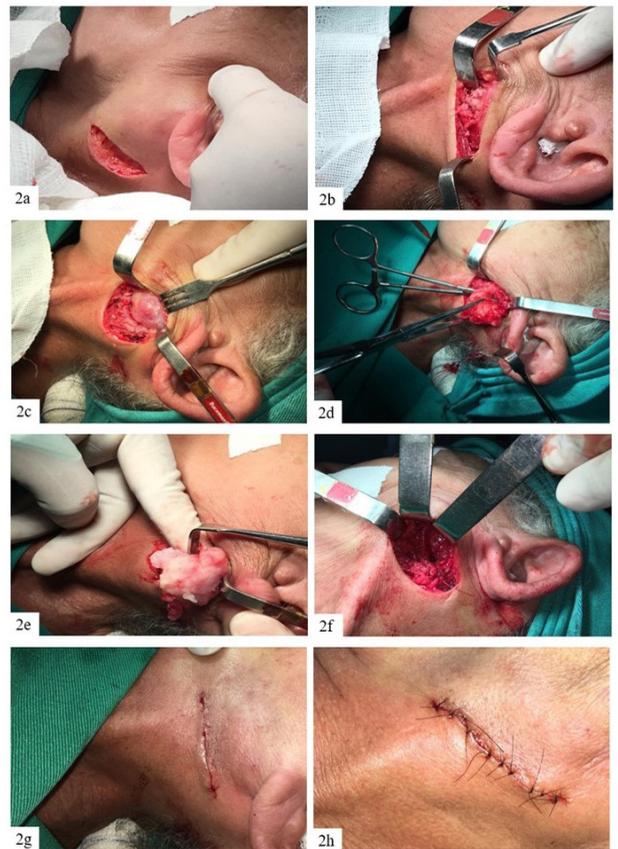


Source: The authors.

Computed tomography revealed a well-delimited, circumscribed hyperdense lesion, approximately 35 mm in its largest diameter, irregular surface, located superficially at the left parotid gland. After clinical evaluation and complementary tests, the diagnostic hypothesis of pleomorphic adenoma was raised.

The surgical planning proposed consisted of excisional biopsy for lesion enucleation. The procedure was performed in a surgical center under general anesthesia balanced by nasotracheal intubation. The access of choice was the submandibular located in the cervical region just below the lesion (Figure 2a and 2b). The divulsion continued until the capsule of the lesion, individualizing it in order to release it from all adherences (Figure 2c). The manipulation and clamping of the lesion caused the leakage of viscous content of whitish staining (figure 2d and 2e). After dissection of the lesion and its excision, hemostasis and copious irrigation of the surgical wound were performed (Figure 2f). The suture was performed by tissue planes, with 4-0 polyglycolic acid absorbable thread in the internal planes and the skin suture with nylon 5-0 (Figure 2g and 2h).

**Figure 2** - Surgical procedure steps. (2a and 2b) Conservative submandibular access; (2c) Lesional capsule disclosure; (2d and 2e) Lesion individualization and clamping; (2f) Surgical bed after lesion removal; (2g) Internal flat suture; (2h) Skin sutures



Source: The authors.

Lesion analysis showed the presence of capsule with whitish and viscous content inside (Figure 3a), size of approximately 35 mm, stony consistency and rounded shape (Figure 3b). The lesion storage was performed in 10% formaldehyde solution and the part was referred for anatomic-histopathological evaluation.

**Figure 3** - Lesion after removal of surgical bed. (3a) Whitish lesion after removal of the surgical bed; (3b) Approximate size of 3.5 cm in its largest diameter



Source: The authors.

In the seven-day postoperative period, the skin sutures were removed, a good scar aspect was observed without evidence of dehiscence, cardinal signs of inflammation or infection (Figure 4a and 4b). The mimicry motricity was preserved and the patient did not present complaints (Figure 4c and 4d).

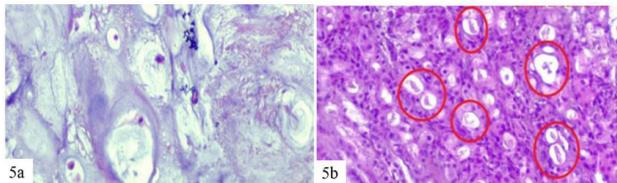
**Figure 4** - Evolution of 7 days surgical procedure. (4a) 7-day postoperative period, skin sutures with good scar aspect are noted; (4b) In evidence, suture area with excellent co-opted area and absence of signs of inflammation; (4c and 4d) Motricity of preserved mimicry is noted



Source: The authors.

The histopathological report confirmed the diagnostic hypothesis of pleomorphic adenoma (Figure 5a and 5b).

**Figure 5** - Confirmatory histopathological examination of pleomorphic adenoma. (5a) The histological sections reveal a fragment of ectodermal neoplasm originating from the salivary gland represented almost exclusively by chondroid tissue; (5b) Histological sections reveal a fragment of ectodermal neoplasm originating from salivary gland, consisting of solid parenchyma permeated by a few small ductiform structures composed of two cellular layers interpreted as being: neoplastic ductal epithelial cell and neoplastic myoepithelial cell. Extensive chondroid areas are also observed. The neoplastic stroma is formed by fibrous connective tissue



Source: The authors.

**Figure 6** - Patient after six post-operative months period.



Source: The authors.

In the image, there were no signs of recurrence at the operated site.

## 2.1 Discussion

Pleomorphic adenoma is the most common benign neoplasm affecting the parotid gland<sup>2,15</sup>. It presents a predilection for the female gender in a variable proportion found in the literature, with an average of around 1,5:1, and although it can be found in all ages, it occurs more frequently from the 4<sup>th</sup> to 6<sup>th</sup> decade of life<sup>6,9,20</sup>. The clinical case described in this study corroborates the epidemiological profile of the patients described in the literature.

In a retrospective clinical study of 68 patients operated on with diagnosis of pleomorphic adenoma of parotid gland, the authors observed a higher frequency of pleomorphic adenoma in the left parotid gland, 61.5% of the cases, with a ratio of 1.6:1 when compared to the right side. The clinical case reported also presents location on the left side, however, in the literature there is currently no consensus regarding the side of involvement of these lesions, therefore, requiring further clarification regarding this aspect in future studies<sup>9</sup>.

Although salivary glands are considered by some authors to be relatively simple organs, the same cannot be asserted for neoplasms that affect them, mainly due to their rich histological variety<sup>21</sup>. PA also called "mixed tumor" due to its histological diversity is the most frequent benign salivary gland neoplasm in humans. In essence, they are tumors derived from myoepithelium, with both epithelial and mesenchymal differentiation<sup>3,4</sup>.

Due to the fact that PA presents itself as a slow and asymptomatic growth lesion, it often leads to late demand for medical care and diagnosis of lesion<sup>5,6</sup>. The great majority (90%) of pleomorphic adenomas affect the superficial lobe of the gland, being 80% in its lower portion<sup>22</sup>. Facial motor involvement is not common in cases of pleomorphic adenoma of parotid gland, although it may occur by compression or stretching of the nerve by voluminous tumors<sup>23</sup>.

Image exams, although not essential, are extremely important, since they help to determine the location, extent and shape of the lesion, contributing to the complementation of the diagnosis and to surgical planning. Salivary glands pathologies present several image characteristics and the examination to be chosen depends on some factors, such as the lesion clinical presentation, consistency, size and location<sup>9,11,24</sup>.

Among the most widely used, magnetic resonance imaging (MRI) is ultrasound (USG) and computed tomography (CT), usually indicated for help in the diagnosis of soft and bone tissue lesions. The ultrasound examination assists in the assessment of lesion consistency<sup>11,24</sup>, however, the magnetic resonance imaging is particularly useful to demonstrate the interface of tumor and surrounding tissues<sup>15</sup>. Pleomorphic adenoma often has a characteristic appearance in magnetic resonance images, presenting homogeneous hyperintensity

in T2, well-circumscribed borders and solid contrast enhancement<sup>24-26</sup>.

As for the treatment form, the lesions located in the superficial lobe of the parotid gland are better treated through superficial parotidectomy, with identification and preservation of the facial nerve. On the other hand, lesions located in the deep lobe, total parotidectomy is usually necessary, if possible, with the preservation of the facial nerve. Local enucleation should be avoided, as the neoplasm may not be completely removed or the capsule may be violated, resulting in cells remaining in the tumor bed. Conservative enucleation can result in recurrence, making it difficult to manage these cases due to the multifocal dissemination in the primary bed<sup>11,12</sup>.

The exact causes of recurrence of pleomorphic adenoma remain contradictory. The widely accepted hypothesis is the subtotal removal of the tumor due to inadequate surgery, while the characteristics of the tumor capsule or other histological aspects are rarely examined<sup>12,27</sup>. Recurrence rates range from zero to 14% when the surgical procedure is extra-capsular dissection and the main cause for this recurrence is incomplete surgical resection of the pleomorphic adenoma capsule due to partial exposure or rupture in the transoperative period<sup>14,27,28</sup>.

Investigating the recurrence rate of pleomorphic adenoma in the evaluation of 47 patients with at least one episode of tumor recurrence, 41 cases were in parotid gland. They reached the conclusion that primary tumor surgery included tumor removal in only 36% of cases. Despite the surgical method, all surgical techniques for removing the pleomorphic adenoma of the superficial parotid exposes the tumor capsule focally. The study was unable to determine tumor effusion after rupture of the primary tumor capsule, and it is only known in some cases. Rupture of the primary tumor capsule and tumor effusion are strongly associated with recurrence. However, the authors found that rupture of a capsule does not always lead to recurrence and there are several reports that did not show association of sowing of tumor cells with recurrences<sup>13,28,29</sup>.

Advanced reasons for the recurrence of pleomorphic adenoma can be grouped into pathological (capsule thickness, lack of capsule, pseudopod, satellite nodules) and related to surgery (tumor rupture, tumor content spill, insufficient resection margins due to nervous branches, inadequate excision related to the type of surgery). The authors found that recurrence seems to be increased in the subtype of myxoid pleomorphic adenoma in which the capsule is often thinner and incomplete. Surgical variables related to recurrence include positive margins and tumor effusion, as already described by other authors. Nevertheless, the primary treatment of pleomorphic adenoma should be carefully planned, performed and centered on the complete lesion removal<sup>13,17,18,27,28</sup>.

The most common complication in the treatment of parotid pleomorphic adenoma is facial nerve involvement. In the present case, there was no functional impairment of the nerve, probably due to the technique used, extra-capsular dissection, which has lower morbidity to the nerve

in question. According to Tiago & Suzano, 2003 paresis of the facial nerve is 2.3 times greater when total parotidectomy is performed, when compared to superficial parotidectomy. In addition to facial nerve involvement, another complication that can be found is Frey's Syndrome, which is clinically presented with sweating and erythema in the parotid region, caused by interruption of parasympathetic post-ganglionic fibers of the glossopharyngeal nerve that travel through the auriculotemporal nerve that innervates the parotid region<sup>10,19,28</sup>.

Although this is a benign lesion, with low recurrence rate and malignant transformation<sup>15</sup>, periodic monitoring for a minimum period of 05 years is important. Currently, the patient is in six months postoperative period, with no signs of recurrence.

#### 4 Conclusion

Although this is a benign tumor, the previous diagnosis and treatment of pleomorphic adenoma is important, because when treated early, it has a favorable prognosis and a lower chance of recurrence. Although the possibility of recurrence is small, the patient should be monitored and evaluated periodically for at least five years. The patient of the clinical case described is in a six-month postoperative period with good evolution, with no sign of recurrence. Therefore, it can be concluded that the treatment so far is efficient and effective.

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