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

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

Davisson Alves Pereira*ab; Eduvaldo Campos Soares Juniorab; Mariana da Silva Bonattoab; Kallyne Munik Souza Moratoab; André Luis Fernandes da Silvab

*Federal Universidade of Uberlândia, Stricto Sensu Post-Graduate Program in Dentistry. MG, Brazil. 
abUniversity of Cuiabá, Residence Program in Surgery and Buccomaxillofacial Traumatology. MT, Brazil. 
E-mail: davisoncaneda@hotmail.com

Recebido em: 31/01/2020 
Aprovado em: 04/05/2020

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 of 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 at reporting a case of pleomorphic adenoma of the parotid gland with eight years of evolution. The tumor was treated conservatively. Patient has been followed for six months, and no sensory alterations, fistulae, clinical signs of recurrence, nor any other alterations have been observed so far. Due to the lesion benign nature, an evolution with favorable prognosis is expected.

Keywords: Adenoma. Neoplasms. Salivary Glands.

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 region1-2. SGTs have an annual incidence in the west of from 2.5 to 3.0 cases for every 100,000 persons 3-7 .

Within this group, pleomorphic adenoma (PA), the most common benign neoplasm of the major salivary glands (GSMA) and minor ones (GSMe) stand out3,8,9. This lesion is described as a slow evolution neoplasm, well delimited, painless, apparently encapsulated, that rarely exceeds 5 cm in its largest diameter3,5,10 . The PA capacity to undergo recurrences and malignant transformation has been reported by the 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 treatment11-15.

Although the clinical and histological aspects of this neoplasia are well known, its etiology and pathogenesis remain uncertain4 . However, it is known that in more than 70% of the cases there is an association with abnormal karyotypes, involving a locus of the PLAG1 gene (Pleomorphic adenoma gene 1). Recently, new genes that may be involved in tumor formation have been identified16. In addition, there are evidences that exposure to UV (ultraviolet) rays increases the risk of developing it3.

The treatment of choice for this neoplasia consists of complete surgical excision17. 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 recommended7,14,18,19. 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, 66 years old, brown, housemaid, appeared at the outpatient clinic of the Buccomaxillofacial
Surgery and Traumatology Service of General Hospital of Cuiabá – MT, presenting a volumetric increase on the left side face, with evolution time 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 extra-oral physical examination, a mobile volumetric increase was observed in the left parotid gland region of approximately 35 mm in its largest diameter, sessile basis, 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.

Computed tomography revealed a well-delimited, circumscribed hyperdense lesion, approximately 35 mm in its largest diameter, irregular surface, located superficially in 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 one located in the cervical region just below the lesion (Figure 2a and 2b). The division continued until the capsule of the lesion, individualizing it in order to release it from all the adherences (Figure 2c). The manipulation and clamping of the lesion caused the leakage of whitish staining viscous content (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 suture of the skin 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.

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

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

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

Figure 4 - Evolution of 7 days of surgical procedure process. (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) Motor movement of preserved mimicry is noted

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. 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 4th to 6th decade of life. 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 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.

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. The PA also called "mixed tumor" due to its histological diversity is the most frequent benign salivary gland neoplasia in humans. In essence, they are tumors derived from myoepithelium, with both epithelial and mesenchymal differentiation.

Due to the fact that PA is a slow and asymptomatic growth lesion, it often leads to late demand for medical care and diagnosis of lesion. The great majority (90%) of pleomorphic adenomas affect the gland superficial lobe, being 80% in its lower portion. 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.

Image exams, although not essential, are extremely important, since they help to determine the location, extent and shape of the lesion, contributing to the diagnosis complementation 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.

Among the most widely used, magnetic resonance imaging (MRI) is ultrasound (USG) and computed tomography (CT), usually indicated for support in the diagnosis of soft and bone tissue lesions. The ultrasound examination assists in the assessment of lesion consistency, while the magnetic resonance image is particularly useful to demonstrate the tumor and surrounding tissues interface. Pleomorphic adenoma often has a characteristic appearance in magnetic resonance images, presenting homogeneous hyperintensity in T2, well-
circumscribed borders and solid contrast enhancement\textsuperscript{14,26}.

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 facial nerve preservation. 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\textsuperscript{11,12}.

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\textsuperscript{12,27}. Recurrence rates range from zero to 14\% when the surgical procedure is extracapsular dissection and the main cause for this recurrence is the incomplete surgical resection of the pleomorphic adenoma capsule, due to partial exposure or rupture in the trans operative period\textsuperscript{14,27,28}.

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 expose the tumor capsule focally. The study was unable to determine tumor effusion after rupture of the primary tumor capsule, and 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 tumor cells seeding with recurrences\textsuperscript{13,26,29}.

Advanced reasons for the pleomorphic adenoma recurrence 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 mixed pleomorphic adenoma in which the capsule is often thinner and incomplete. Surgical variables related to recurrence include positive margins and tumor shedding, as already described by other authors. Nevertheless, the primary treatment of pleomorphic adenoma should be carefully planned, performed and centered on the lesion complete removal\textsuperscript{13,17,18,27,28}.

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 et al.,\textsuperscript{9} facial nerve paresis 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 parasym pathetic-post-ganglionic fibers of the glossopharyngeal nerve that travel through the auriculotemporal nerve that infuses the parotid region\textsuperscript{10,19,28}.

Although this is a benign lesion, with low recurrence rate and malignant transformation\textsuperscript{15} periodic follow-up is important for a minimum period of five years. Currently, the patient is in six months postoperative period, with no signs of recurrence.

3 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 low, the patient should be monitored and evaluated periodically for at least five years. The patient of the described clinical case 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.

References

7. Speight PM, Barrett AW. Salivary gland tumours. Oral