Giant pleomorphic adenoma of the parotid gland

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Abstract
Pleomorphic adenoma is the most common type of all benign and malignant salivary gland tumors, involving more frequently the parotid gland. It is a benign tumor with a slow and continuous growth that without treatment can reach an enormous size. We present a case of a giant pleomorphic adenoma in a 78-year-old man with a history of more than 30 years of a growing lesion in the parotid gland. Clinical examination revealed a giant mass on the right side of the face, however without any sign of facial nerve damage. The tumor was completely resected by total parotidectomy and preservation of the facial nerve. Macroscopically, the tumor measured 28 cm and weighed 4.0 Kg. On the histological examination there was a predominance of epithelial and mioepithelial cells in a hyaline and mixoid stroma. It was not found any area of malignant transformation. In the post-operative the aesthetic and functional results were excellent.

Key words: Pleomorphic adenoma, parotid.

Introduction
About 70% of all salivary gland tumors arise in the parotid gland, and approximately 85% are benign. Pleomorphic adenoma (PA) represents 45-74% of all salivary gland tumors and 65% of them occur in the parotid gland.(1,2,3) PA presents clinically as a painless, slow-growing mass, usually varying from 2-6 cm when resected.(4) Cases of giant PA have been reported in the parotid gland, presenting as an irregular multinodular mass that can weigh more than 8 Kg.(1) Most cases of giant PA were seen before 1980’s, but some cases have been published recently.(4-5) These unusual cases are treated by parotidectomy, but the possibility of positive surgical margins and malignant changes must be considered.

This paper describes a case of a giant pleomorphic adenoma arising in the parotid gland and treated by total parotidectomy with facial nerve preservation.

Case report
A 78-year-old man presented to the Department of Head and Neck Surgery and Otorhinolaryngology, Hospital do Cancer A.C. Camargo, São Paulo, Brazil, complaining of a tumor on the right side of the face for more than 30 years. The patient lived alone, isolated from his family and did not present any systemic disease. Clinical examination showed a giant, firm, multinodular, irregular and painless mass on the right side of the face crossing the mid-line, measuring approximately 30 cm of extension, involving the parotid and cervical regions (Figure 1). Despite of the great dimension of the tumor, there were no signs of facial nerve palsy. The skin that covered the lesion presented some ulcerated areas. The main clinical hypothesis of diagnosis was a benign tumor of the parotid gland, probably pleomorphic adenoma. Under general anesthesia, it was performed a right total parotidectomy with preservation
Giant pleomorphic adenoma of the facial nerve and complete removal of the tumor en bloc. The postoperative course was uneventful. Macroscopically, the removed mass measured 28cm x 20cm x 16cm, and weighed 4.0 Kg. Microscopically the tumor was composed of islands and strands of epithelial cells immersed in a hyaline stroma, some showing squamous differentiation. Areas presenting spindle and plasmocytoid myoepithelial cells in a myxoid stroma were also abundant (Figure 2). All pieces of the tumor were microscopically analyzed and none of them showed evidences of malignant changes. The final diagnosis was of pleomorphic adenoma with negative surgical margins. The patient presented excellent aesthetic and functional results, without signs of facial nerve palsy or recurrence (Figure 3).

Discussion

Pleomorphic adenoma is the most common salivary gland tumor. The main site of occurrence is the parotid gland, affecting patients of any age, more frequently between the fifth and sixth decades of life. Although uncommon, cases of giant PA have been reported, most of them involving the parotid gland. In 1989, Schultz-Coulon reviewed 31 cases of giant PAs of the parotid gland. He found a female predominance (64.5%), with an age range of 20 to 40 years old, and weight of the tumor between 1 to 27 Kg. Buenting et al. (4) (1988) reviewed the ten largest pleomorphic adenomas ever published in the English-language literature, most of them before the 1980’s. They found a mean tumor weight of 7.81 Kg, nine out of ten cases occurred in female, with a mean age of 56.2 years. In our case, the patient was a man who presented a tumor with history of 30 years of evolution and weighed 4 Kg, similar to more recent reports. In most of the cases described it is considered that lack of information and negligence of the patients is relevant for the long course of an evident clinical mass, and in our case it must be considered that the patient lived alone and isolated from his family.

Microscopically, PAs are characterized by a myriad of morphological diversity. Epithelial cells are arranged in sheets and islands showing typical ductal structures, and various epithelial and myoepithelial characteristics as spindle, clear, squamous, basaloid, plasmacytoid, oncocytic and sebaceous. The stroma characteristically is mixed, with fibrous, chondroid, myxoid or hyaline aspects. The current case was formed predominantly by cuboidal and spindled cells embedded in a hyaline and myxoid stroma.

The incidence of malignant transformation in PAs ranges from 1.9% to 23.3%. The risk increases in tumors with long time of evolution, recurrences, advanced age of the patient and location in a major salivary gland. Some authors postulated that the risk of malignant transformation increases from 1.6% in tumor with less than 5 years of evolution, to 9.5% for those presenting for more than
15 years.(10) The classic clinical history of carcinoma ex-pleomorphic adenoma is of a slow-growing mass for many years, with a recent fast growth.(11) A case of a giant PA with malignant transformation with this typical history was recently reported by Honda et al.(5) (2005) in a 72-year-old woman with a slow growing parotid lesion for 20 years, with a rapid increase in the last 3 months. Schultz-Coulon (6) (1989) reported 31 cases of giant PAs, and in 3 cases malignant areas were found within the tumor. Our patient presented all the characteristics for an increased risk of malignancy, however either clinically and microscopically there were no such evidences.

The treatment of choice for PAs of the parotid gland is superficial parotidectomy with preservation of the facial nerve.(12) In two recent cases of giant PA reported in the literature, the resection of the tumors were performed with preservation of the facial nerve.(5) Our case was treated by total parotidectomy because of the tumor size, and the facial nerve was preserved, with excellent aesthetic and functional results. Currently, giant parotid tumors are rarely seen, but they still occur. Patient’s lack of information and also fear of surgery seems to be the main reasons for these long standing lesions. Nevertheless, if there is no malignant transformation usually giant PAs are completely resected with excellent results and minimal side effects.

References