

Parotis Bezinin Tekrarlayan Çok Odaklı Pleomorfik Adenomu, Olgu Sunumu

Recurrent Multifocal Pleomorphic Adenoma of the Parotid Gland, A Case Report.

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ÖZ

Pleomorfik adenoma, parotis bezinin benign tümörleri arasında en yaygın olanıdır. Tedavisi cerrahidir. Enükleasyon ameliyatı yapılanlarda rekürrens oranı yüksektir. Rekürrensleri en aza indirmek için fasiyal sinirin korunduğu yüzeysel parotidektomi ameliyatı yapılmalıdır. Rekürrenslerde tedavi, total parotidektomidir, rekürrensler tek nodül veya çok odaklı olabilmektedir. Tekrarlayan çok odaklı pleomorfik adenomlarda en önemli tanı yönteminin Manyetik Rezonans Görüntüleme (MRG) olduğu bilinmektedir. Bu makalede parotis bezinin tekrarlayan, çok odaklı pleomorfik adenom olgusunu sunuyoruz.

Anahtar Kelimeler: Tekrarlayan çok odaklı pleomorfik adenoma, Parotis bezi tümörü, Total parotidektomi.

ABSTRACT

Pleomorphic adenoma is the most common benign tumor of the parotid gland. The treatment is surgery. The rate of recurrence is high when the enucleation surgery is performed. In order to minimize the recurrences, a superficial parotidectomy by protecting facial nerve should be performed. Treatment for recurrences is total parotidectomy, recurrences may be single nodule or multifocal. Magnetic Resonance Imaging (MRI) is known to be the most important diagnostic method for recurrent multifocal pleomorphic adenomas. In this article we present a recurrent multifocal pleomorphic adenoma of the parotid gland.

Key words: Recurrent multifocal pleomorphic adenoma, Parotid gland tumor, Total parotidectomy.

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INTRODUCTION

Pleomorphic adenoma represents 60 - 70 % of all parotid gland benign neoplasms. Pleomorphic adenoma occurs in individuals in most common 4th-5th decades and is seen more often in females than males. Pleomorphic adenoma usually presents as a slow-growing, painless mass and not effects the facial nerve functions (1). Pleomorphic adenoma exhibits wide sitomorphologic diversity and consists of the epithelial and the myoepithelial cell components. Surgical excision is the treatment method. However the rupture of the tumoral mass during the surgery, inadequate excision, injury of the pseudocapsul and inadequate resection of pseudopods increase the recurrence rates. It is hard to treat the recurrent pleomorphic adenoma cases. The revision surgery of parotid gland area has a high morbidity rate. The postoperative adhesions after initial surgery and the anatomical variations causes a 15-30 % rate of risk of injury to the facial nerve (2,3). Recurrence rate varies as, 20-45 % after simple enucleation, 1-4 % after superficial parotidectomy and 0-0.4 % after totally parotidectomy. In most studies it represent that recurrence might have a number from single to hundred however the MRI views and microscopic findings have not a certain number (4,5). In this article we present the case with a number of 23 tumoral focus with sizes vary from 1mm to 50 mm

CASE REPORT

23-year-old male patient presented with a swelling area at the left side of the face, pain and numbness in the same region for 6 years. Physical examination, the left parotid gland examined swelled diffusely and differentsizes of nodules palpated at the same area, facial nerve functions were properly. 8 years prior to this presentation, the patient had undergone a surgery but the pathologic findings report and the epicrisis could not be reached.

The patient complained of swelling, pain and tension in front of his left ear after 24 months of his first operation and he applied to our clinic on the increase of his complaints. Then a parotid gland ultrasonography was examined and whereas different-sized solid nodules detected in this area, a parotid gland MRI had be needed. MRI pointed to

the multifocal lesions in the left parotid gland area, the largest lesion measured approximately 20x24x52 mm and was shown to be isointense on the T1 weighted image (axial), hyperintense on the T2 weighted image (axial and sagittal). The contrasted series showed, medium level contrast retained images (**Figure 1**).

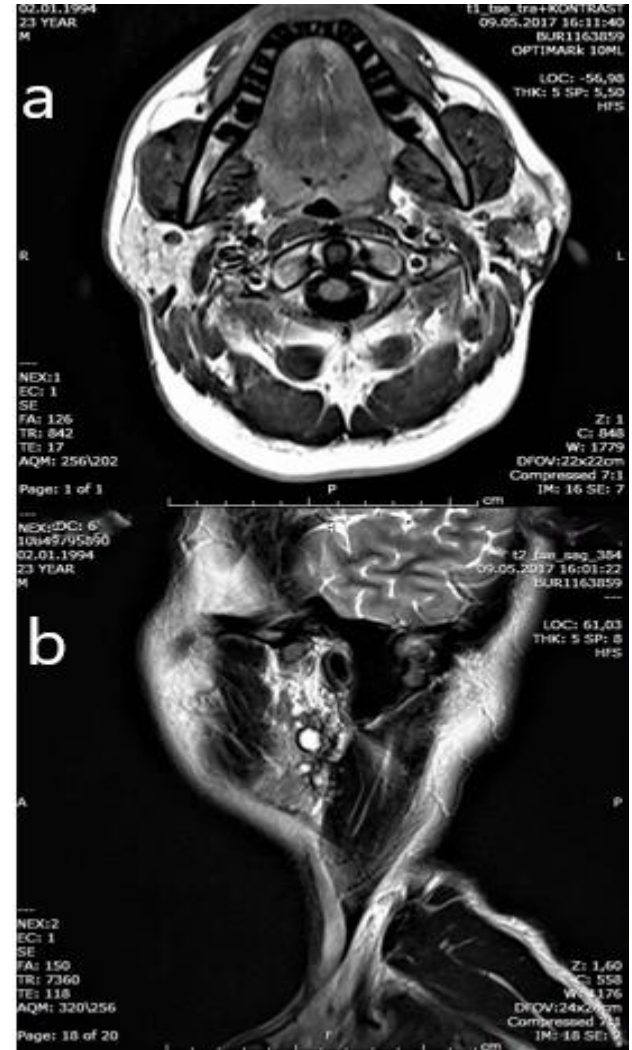


Figure 1. The left parotid gland MRI section, multiple pleomorphic adenoma: a. T1-weighted isointense (axial). b. T2-weighted hyperintense section(sagittal)

Ultrasonography guided Fine-needle aspiration biopsy was performed and specimen was sent for histopathologic investigations. These findings were reported as pleomorphic adenoma. At the date of 16.05.2017 a modified blair incision with the scarring tissue in the left preauricular region included to the specimen was performed.

A microscop guided total parotidectomy surgery performed with an attention to the facial nerve protection and all of the tumoral lesions were removed and included to the specimen. Perioperative photos of the multifocal tumoral lesions and the facial nerve was taken (Figure 2).

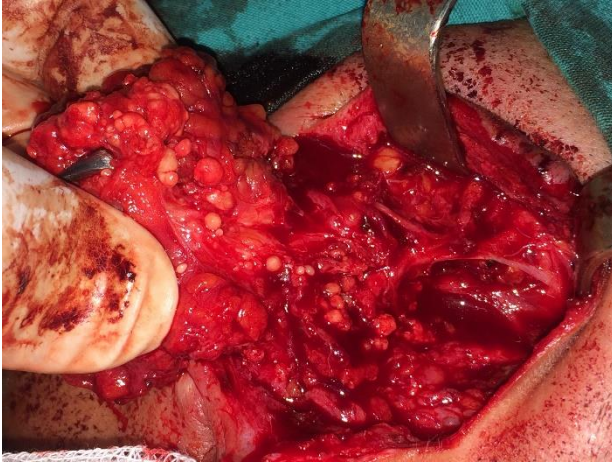


Figure 2. Perioperative photography, the view of the facial nerve and the different sized tumor focuses

The pathologic material of this total parotidectomy surgery showed an amount of 23 different tumoral focus of pleomorphic adenoma. The sizes differed from 1 mm to 50 mm. Malignant transformation was not detected (**Figure 3**).

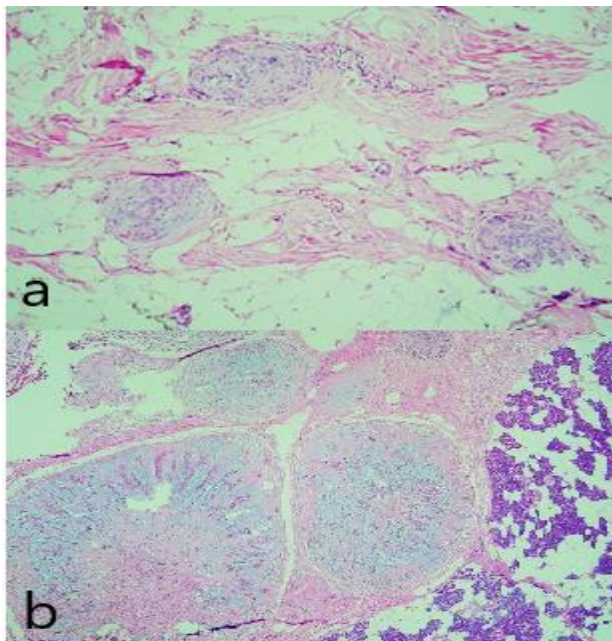


Figure 3. Histopathology of the tumor: a. A nodular patterned stroma included tumoural area with the neighborhood of the benign parotid gland (x40 H&E). b. Microscopic tumour focuses scattered in the surrounding adipose tissue (marked with arrow) (x100 H&E).

Written patient's consent: Written consent was obtained from the patient participating in this study.

DISCUSSION

It is generally accepted that the standart treatment of pleomorphic adenoma is superficial parotidectomy with the preservation of the facial nerve and the removal of the tumor with a surrounding cuff of normal parotid gland tissue (2,3,5). However the treatment of reccurent pleomorphic adenoma is debated, treatment options varies from observation only, adjuvant radiotherapy and to the facial nerve sacrificed radical parotidectomy. To the contrary of this all treatment modalities, the reccurence rate is 10-63% and the certain treatment modal is debated (4,6).

The first hypothesis about the recurrent pleomorphic adenomas was reported in 1950's by Patey and Thackray and it was the microscopic pseudopods that protruding beyond the pseudocapsules of the tumor (7).

It is believed that due to the excision closely to the pseudocapsules, causes inadequate resection of the pseudopods increase the risk of reccurent pleomorphic adenoma (3). Another reason is the rupture of the pseudocapsule thus tumor spillage to the surgical area. Multifocal tumors seen in the incision scarring area and the follow up findings of tumor ruptured surgery patients, that related of increased reccurency, encouraged the hypothesis (6).

Riad and his friends studies showed that, the rupture of the psudocapsule is a significant risk factor of the recurency and tumor spillage. The size of the tumor and the involvement of the facial nerve is important to psodocapsule rupture risk (2). During enucleation the rupture of the pseudocapsule causes new tumoral areas in the surgery field and many tumoral areas outer of the surgery field (1,3).

It is also possible that sometimes initial surgery starts with primary multifocal tumors. Batsakis in 1979 reported that pleomorphic adenoma can be seen primary multifocal in a salivary gland tissue (6). Egmond et al. (8) reported a case in only one parotid gland, which had a primary multifocal

pleomorphic adenoma, with the number of 15 nodules, size varied from 1 mm to 16 mm.

However many studies showed that hundreds of tumor focus is seen in recurrence cases, the scanning methods, photography and microscopic findings did not give a certain number (1,6).

To diagnose the recurrent pleomorphic adenoma MRI is preferred because of its ability to show the superficial and deeply lobes to see the multifocal lesions. The tumor has low signal intensity on T1-weighted series and high signal intensity on T2-weighted series. The contrast series show a medium-level retention (9). Despite this, some tumor focuses can not be detected by an MRI. These tumor focuses generally is seen by the surgeon during the operation with the help of microscopic augmentation and mostly identified in specimen by the pathologist (5).

The pleomorphic adenoma case we studied had a multifocal sight. It was pointed out by the MRI sections and had a great role to determine the surgical plan (Fig. 1). Microscop guided and the facial nerve preserved total parotidectomy surgery is performed and the all of the nodular tumor focuses removed (Fig.2). The microscopic determination of the specimen showed 23 different pleomorphic adenoma focus size varies from 1 mm to 50 mm (Fig. 3).

A recurrent pleomorphic adenoma type in which nodular tumor is detected in the deep lobe by the MRI, total parotidectomy surgery should be preferred for this situation. Enucleation surgery do not need dissection of the facial nerve, so that adhesion and fibrosis around the facial nerve not occur (4,6). The case of this article had a microscop guided and facial nerve preserved operation and no fibrosis and adhesion occurred. Therefore it was suggestive of the first surgery kind was enucleation. The facial nerve functioned properly in the postoperative examination.

Many authors, Stennert et al. expressed the total parotidectomy when a recurrent case is seen after enucleation or superficial parotidectomy surgery, to include the multifocal lesions that can be overlooked to resection (10). If tumor is contacted with the facial nerve, the total parotidectomy surgery can not remove tumor totally. During the

dissection of the facial nerve, leaving microscopic residuals and spillage of the tumor can be seen quite likely. At this condition an extended surgery including the affected branches of the facial nerve or radical parotidectomy should be thought. The rate of including the branches of the facial nerve to the resection in the recurrent pleomorphic adenoma cases is 14-30% (3,6). The facial nerve branches was preserved in our case.

Witt et al (6). Emphasized the adjuvant radiotherapy showing that adjuvant radiotherapy increases the success rate of local controlling of the multifocal and the multiple recurrent pleomorphic adenoma cases and if repeating occurs, it can be performed effectively on to the facial nerve injured patients. Andreasen et al (11). reported that a rate of 46.6 % recurrence was seen in 148 recurrent pleomorphic adenoma cases even though surgery and adjuvant radiotherapy was performed. Maxwell et al (12). reported that they performed total parotidectomy to the 35 patients with recurrent pleomorphic adenoma diagnosis, and did not give radiotherapy, as a result they had a 23% rate of recurrence.

The MRI have a significant role to determine the pleomorphic adenoma if it origins primary multifocal or not, and also for identification of recurrent multifocal pleomorphic adenoma and to planning the surgery. Microscopic method is the final diagnostic method. The acceptable treatment method of recurrent pleomorphic adenoma is facial nerve preserved total parotidectomy, but performed especially by an experienced surgeon and it needs an attention to the multifocal structure and to the connection between the facial nerve and the tumor. However if tumor is in contact with the facial nerve, the nerve can be sacrificed. The postoperative follow ups of this patients, can prevent probable additional recurrences at earlier periods.

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