Synchronous Tumors of the Parotid Gland: Presentation of 3 Cases and a Review of the Literature

Parotis Bezin Senkron Tümörü: Üç Olgu Sunumu ve Literatür Derlemesi

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ABSTRACT

Synchronous tumors of the parotid gland are two or more neoplasms originating from different sites of the gland, making up less than 1% of all parotid tumors, most frequently as Warthin tumors at more than one site. In this study, 3 cases of synchronous parotid tumors out of 193 parotid surgeries performed between 2004-2010 in our clinic will be presented with their clinical, radiological and histopathological findings, and discussed according to the current literature. The first case was diagnosed as synchronous parotid tumor in composition of pleomorphic adenoma and Warthin tumor. The second case was Warthin tumor at 2 different sites, and the third case was a salivary duct carcinoma with Warthin tumor. Synchronous tumors, despite being rare, are remarkable as they can affect the prognosis and should be kept in mind during the differential diagnosis of parotid tumors.

Key Words: Synchronous tumor, Warthin tumor, pleomorphic adenoma, salivary duct carcinoma, parotid tumor, differantial diagnosis

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ÖZET

Parotis bezi senkron tümörleri bezin farklı kısımlarından gelişmiş iki veya daha fazla neoplazmla karakterizedir. Parotis bezi senkron tümör sıklığı bezin tüm tümörlerinin %1'inden daha az kısmını oluşturmaktadır ve sıklıkla birden fazla odakta Warthin tümörü şeklinde görülmektedir. Bu çalışmada kliniğimizde 2006-2010 arasında parotis cerrahisi yapılmış 193 hasta içinden senkron parotis tümörü saptanan 3 vaka, klinik ve histopatolojik bulguları eşliğinde tartışılmıştır. İlk vaka pleomorfik adenom ve Warthin tümöründen oluşan senkron tümördür. İkinci vakada 2 odakta Wartin tümörü saptanmıştır. Üçüncü vakada ise tükürük bezi duktal karsinomu ve warthin tümörü birlikteliği saptanmıştır. Senkron tümörler nadir olsalar da prognozu etkileyebilecekleri için önemlidir ve parotis tümörlerinin ayırıcı tanısında akılda tutulmalıdır

Anahtar Sözcükler: Senkron tümör, Warthin tümörü, pleomorfik adenom, türük bezi kanalı tümörü, parotis tümörü, ayırıcı tanı

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INTRODUCTION

Salivary gland tumors make up 3% of all head and neck tumors. 75% of these neoplasms are located in major salivary ducts, most frequently in the parotid gland (1). Synchronous tumors of the parotid gland are two or more neoplasms originating from different sites of the gland (2). Unilateral or bilateral synchronous tumors make up less than 1% of all parotid tumors (3). Synchronous tumors of different histological types are less than 0.3% of all major salivary gland tumors (4). The coexistence of benign and malign tumors is even rarer. In this study 3 cases of synchronous parotid tumor are presented.

CASE REPORT

Case 1

Sixty-eight-year-old male patient presented with a painless swelling below his left ear lobule, which had been present there for 3 months. In medical history, it was noted that he had vertical larynjectomy laryngectomy and adjuvant 6000 grey radiotheraphy for laryngeal squamous cell carcinoma 26 years ago. No recurrences were observed in his controls, and he was considered to be cured. He also had a history of smoking for 25 years with approximately 20 cigarettes a day.

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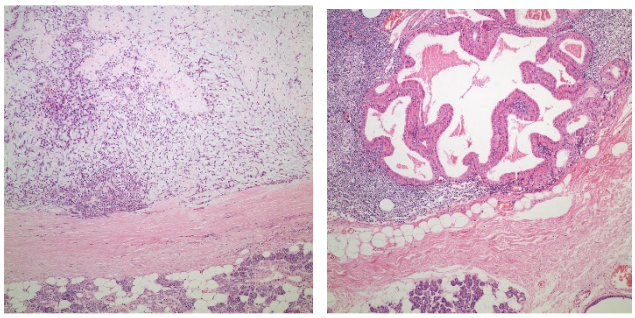
Physical examination showed a firm, mobile, non-tender 2x2 cm mass with regular margins below his left ear lobule. Magnetic resonance imaging (MRI) scans revealed a 15mm, lobulated mass located under the superfical lobe of the left parotid gland. The mass was hyperintense at T1A series, and showed contrast enhancement. Fine needle aspiration biopsy (FNAB) was nondiagnostic. The patient underwent partial parotidectomy. Histopathologically, the main mass was solid-myxoid in appearence, and was diagnosed as pleomorphic adenoma. A focal Warthin tumor was also noted in a different site (Figure 1A, B). After 36 months of follow up, the patient was asymptomatic.

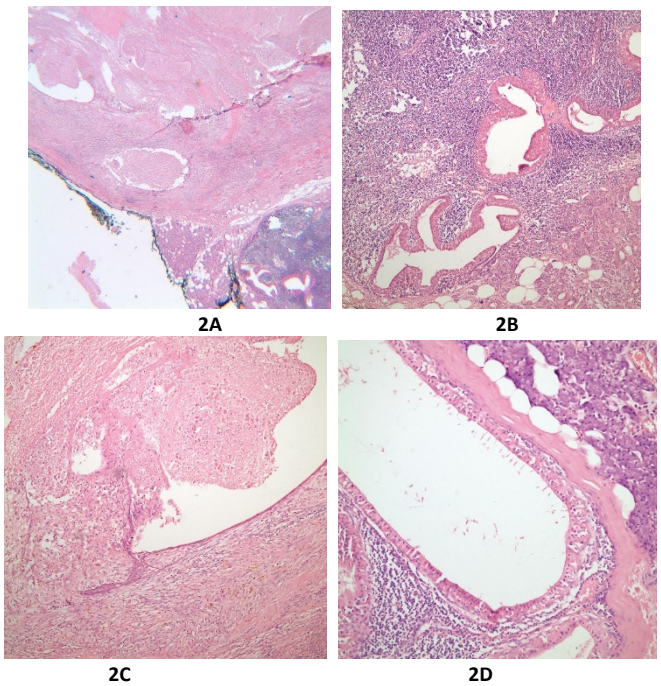


1A 1B

Figure 1: Pleomorphic adenoma (1A) and Warthin tumor (1B): Well circumscribed pleomorphic adenoma, composed of myoepithelial cells and chondroid matrix was accompanied by a small intranodal papillary proliferation of oncocytic cells, diagnostic for Warthin tumor.

Case 2

Fifty-eight-year-old male patient presented with a slow growing painless swelling, which had been present for 2 years beneath his right ear. Apart from the habit of smoking 40 cigarettes a day for 25 years, his medical history was unremarkable. Computerized tomography (CT) revealed a 3x 3x 3.5 cm mass with regular margins and the heterogenious contrast enhancement. FNAB was indicative of a Warthin tumor. The patient underwent right partial parotidectomy. The pathological examination revealed two isolated Warthin tumor lesions. The bigger mass was almost completely infarcted, and had multiple reactive neighbouring lymph nodes (Figure 2A, B, C, D). After 24 months of the follow up the patient was asymptomatic.



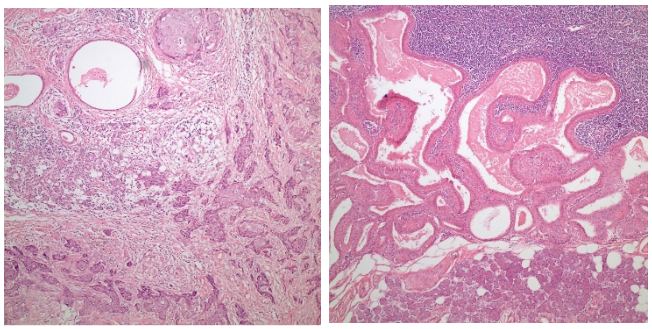
2A 2B

2C 2D

Figure 2: Two distinct Warthin tumors, one of which had totally undergone necrosis (2A) due to a previously done, diagnostic fine needle aspiration biopsy. Small foci of conserved areas (2B) and squamous metaplasia within prominent histiocytic reaction (2C) revealed the presence of Warthin tumor. The second tumor was also composed of 2 layered oncocytic cells forming papillary fronds accompanied by lymphocytes (2D).

Case 3

Fifty-two-year-old female patient presented with painful, firm swelling behind her left ear that had started 2 months earlier. Apart from the habit of smoking 20 cigarettes a day for 25 years, her medical history was unremarkable. MRI scans revealed a lobulated, contrast enhanced, 19 x20 x32 mm mass and a second 12x9x13 mm mass located inferolaterally. Both lesions had the same imaging properties and were hypointense at T1A centrally. Hyperintense and peripherally hypointense at T2A. MRI also revealed multiple lyphadenopathies located at the left upper cervical and posterior cervical chains. Biggest lymphadenopathy was measured 14x10mm, and lymphadenopathies had similar radiological appearences and minimal contrast enhancements. A FNAB of the parotid gland was indicative of a high grade malignant epithelial neoplasia. Partial parotidectomy and neck dissection were planned as the selected treatment modality. During the surgery, a lymph node adjacent to the mass and attached to sternocleidomastoid muscle was encountered. Frozen sections of pathological lymph node was indicative of metastasis of a malignant epithelial tumor. Radical neck dissection was performed accordingly. The Histopathological specimen revealed a salivary duct carcinoma, a focal lesion of Warthin tumor, and 4 metastatic lymph nodes (Figure 3A, B). Widely distributed perineural and lymphovascular invasions were detected. The patient received adjuvant chemoradiotheraphy. In the follow up, local recurrence did not occur but bilateral pulmonary metastases were detected. The patient died due to pulmonary metastase two years after the operation.



3A 3B

Figure 3: Salivary duct carcinoma (3A) and Warthin tumor (3B). A microscopic focus of Warthin tumor within a lymph node was encountered near by a malignant parotid mass, referred as salivary duct carcinoma, characterized by solid squamoid/oncocytic nests and infiltrating trabeculi of malignant cells.

DISCUSSION

Synchronous tumors of major salivary glands make up less than 1% of all major salivary gland tumors (2,3,5). Synchronous tumors originating from different salivary glands are more common than synchronous tumors in the same salivary gland. In our clinic, synchronous salivary gland tumor incidence was 1.55% (3/193) between 2004-2010. Synchronous salivary gland tumors usually consist of tumors of the same histological type and are mostly encountered as multifocal Warthin tumors. Seventeen percent of Warthin tumors are bilateral, and the risk of bilaterality corelates with the amount of nicotine consumption (5). In all of our cases, at least one component of synchronous tumors were Warthin tumor and the patient with unilateral multifocal Warthin tumor had a habit of smoking 40 cigarettes a day for 25 years.

Tumors of different histological types in the same salivary gland are less than 0.3% of all major salivary gland tumors (4). The most common combination is Warthin tumor and pleomorphic adenoma. Two of our cases had tumors of different histological types, and one of them had a Warthin tumor and pleomorphic adenoma combination. Benign and malign synchronous tumors in the same major salivary gland were even rarer. So far, 27 cases have been documented (6). The most common malignant component in synchronous tumors is mucoepidermoid carcinoma. In cases of a benign-malign synchronous tumor, the malign component determines the prognosis. One of our cases had a salivary duct carsinoma and Warthin tumor co-ocurence. Four of 27 documented benign-malign synchronous cases had salivary duct carcinoma as the malign component, and 2 of them were salivary duct tumors and Warthin tumor combinations (6).

There is a lack of consensus about the underlying mechanism of the synchronous salivary gland tumors. But risk factors such as tobacco consumption, old age, radiation exposure, familial predisposition and viral infections (7) affect all of the salivary glands so that the possibility of synchronous tumors should always be taken into account. . All of our cases were over 50 years in age and had a smoking habit. One of them had previously recieved radiotheraphy to the neck.

Preoperative MRI may help diagnosis of synchronous salivary gland tumors, therefore appropriate surgical management can be planned (8). Pathological specimens should always be examined carefully, keeping in mind the possibility of synchronous tumors.

CONCLUSION

In the clinical approach to salivary gland tumors, multifocal tumor risk should always be taken into account. Though rare, synchronous tumors may determine prognosis. Preoperative MRI may help diagnosis of synchronous salivary gland tumors, therefore appropriate surgical management can be determined preoperately. Moreover, diagnosis of a synchronous tumor may mean early diagnosis of a possible malignant component. Pathological specimens should be examined carefully keeping in mind the possibility of synchronous tumors.

Conflict of Interest
No conflict of interest was declared by the authors.

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