

Synchronous Contralateral Benign and Malignant Parotid Gland Tumors: Case Report

Silva IV¹, de Castro Lopes F², Cardoso AR³ and Cardoso JA³

¹Stomatology Department, Centro Hospitalar Vila Nova de Gaia/Espinho, Vila Nova de Gaia, Portugal

²Stomatology and Maxillofacial Surgery Department, Centro Hospitalar do Porto, Porto, Portugal

³Stomatology and Maxillofacial Surgery Department, Portuguese Institute of Oncology of Coimbra, Coimbra, Portugal

***Corresponding author:** Silva IV, Resident, Stomatology Department, Hospital Center of Vila Nova de Gaia/Espinho, Vila Nova de Gaia, Portugal, Tel: +351 915 921 492, E-mail: inesvazsilva@gmail.com

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Abstract

Multiple primary parotid gland tumors are rare, accounting for 1.7-5% of all parotid neoplasms, and the occurrence of bilateral parotid gland tumors is even lower (1.3-3.5%). These tumors can be synchronous or metachronous and the most common histologic type of bilateral parotid gland tumors is Warthin tumor (79.2-90%). Combined benign and malignant lesions and bilateral neoplasms of different histologic types are even less frequently found.

Pleomorphic adenoma, a benign tumor, is the most prevalent salivary gland neoplasm and in the majority of cases occurs in the parotid gland. The risk of malignant transformation increases with time, thus, surgical treatment at the time of diagnosis is ideal.

Mucoepidermoid carcinoma is the most common malignant salivary gland tumor, accounting for 4.2-12% of all parotid gland neoplasms. Low- and intermediate-grade types are less aggressive and are generally cured through complete surgical excision.

Currently, the mechanisms involved in the occurrence of bilateral parotid tumors are still unclear and its development may be coincidental.

The authors report a case of a 46-year-old woman with pleomorphic adenoma and mucoepidermoid carcinoma presenting synchronously on contralateral parotid glands. The patient presented with a mass on the right parotid gland region and bilateral ultrasonography revealed tumors in both parotid glands. Although multiple primary parotid tumors are unusual, it is important to consider that they can occur. Careful preoperative clinical and imagiological evaluation of both parotid glands is necessary whenever a parotid tumor is identified.

Keywords: Parotid Gland; Tumor; Synchronous; Bilateral; Pleomorphic Adenoma; Mucoepidermoid Carcinoma

Introduction

Primary parotid tumors account for approximately 1-3% of head and neck neoplasms [1] and the majority of them is benign (75-85%) [1,2].

Tumoral involvement of the parotid glands is usually solitary but multiple tumors can occur [3-5]. Multiple primary tumors of the parotid gland include bilateral tumors or unilateral multifocal tumors which are primary in nature, excluding recurrent tumors and metastatic lesions [5]. Therefore, these multiple tumors may occur unilaterally or bilaterally and can be synchronous or metachronous [3,5-7].

The occurrence of multiple primary parotid tumors is rare and is reported between 1.7% and 5% of all parotid tumors [3-5]. The incidence of bilateral parotid gland tumors is even lower accounting for 1.3% to 3.5% of the cases [3,5,7,8]. Synchronous bilateral parotid gland tumors are extremely rare [7].

Histologically, these multiple tumors can show identical or distinct features [5,7,8]. Most of bilateral parotid tumors are benign [3,5,6], and Warthin tumor is the most common bilateral parotid gland tumor, accounting for 79,2% to 90% of all cases [4,5]. Combined benign and malignant lesions and bilateral neoplasms of different histologic types are less common [3,5,6]. Regarding other histopathological types of bilateral tumors, pleomorphic adenoma is the second most common. Bilateral acinic cell carcinoma is also well documented [5]. Ethunandan et al., in their study, found that tumors of different histological types accounted for 20% of multiple tumors, and all but one combination included a Warthin tumor [3].

The authors report a case of a 46-year-old woman with pleomorphic adenoma and mucoepidermoid carcinoma presenting on contralateral parotid glands synchronously.

Case report

A 46-year-old woman was referred to the Stomatology and Maxillofacial Surgery Department of the Portuguese Institute of Oncology of Coimbra, with a one-year history of a slow-growing painless mass on the right preauricular area. There were no additional complaints and the patient had no history of trauma, infection, tumors or radiation therapy.

She had previously undergone an ultrasonography of the parotid and neck region bilaterally, which revealed two lesions with 2.7 and 2.0 cm in the posterior-inferior region of right parotid gland as well as a 2.8 cm solid lesion within the left parotid gland.

Upon bilateral parotid examination, two firm non-tender masses were present in the right and left preauricular areas, which were not adhered to the skin or deep tissues. No enlarged cervical lymph nodes were found and there was no evidence of a facial palsy.

Computed tomography of the head and neck (Figure 1) apparently showed several nodules with liquid content within the parotid glands bilaterally (the largest within the right parotid gland with 2.0 cm and the largest within the left parotid gland with 1.6 cm of diameter).

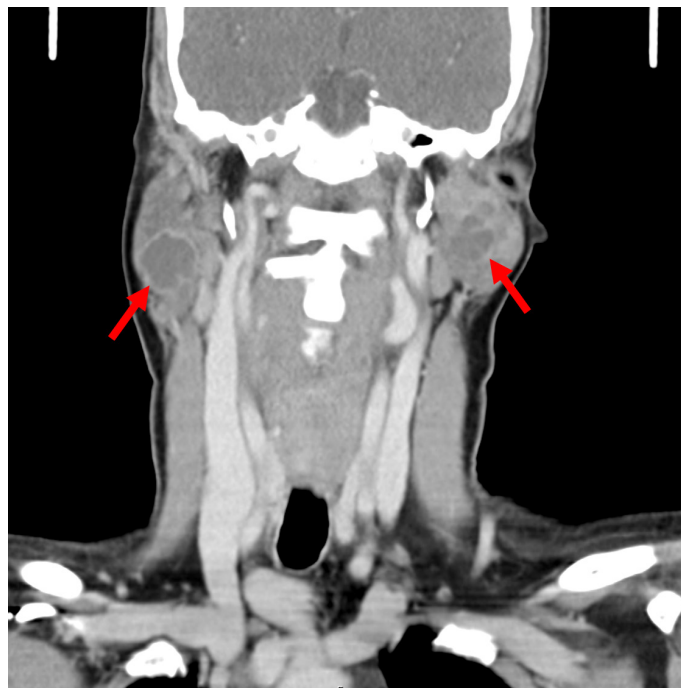


Figure 1: Preoperative Computed Tomography (coronal section) – radiolucent lesions in both parotid glands

Fine needle aspiration biopsy of both sides was inconclusive. Ultrasound-guided core biopsy of the left parotid gland lesion was compatible with the diagnosis of pleomorphic adenoma and on the right parotid gland revealed a cystic lesion, remaining inconclusive regarding the specific diagnosis.

Total parotidectomy was performed bilaterally, in one surgical setting, using the standard retrograde facial nerve approach. Facial nerves branches and stems on both sides were found not to be macroscopically infiltrated by the tumors and were safely preserved.

The histopathological examination revealed a low grade mucoepidermoid carcinoma with 2.1 x 1.5 cm of the right parotid gland and a pleomorphic adenoma of the left parotid gland with 3.0 x 2.0 cm. Both tumors were completely removed, and the lymph nodes included on the specimens did not show neoplastic lesions.

Postoperatively, mild neuropraxia of the facial nerves was found bilaterally and was completely recovered upon the six-month postoperative clinical follow-up.

At the time of this report, ten months post-surgery, there were no signs of recurrence.

Discussion

Salivary gland tumors are rare, ranging from 3% to 6% of all head and neck tumors. Roughly 80% of these present in the parotid gland [9]. The considerable diversity of neoplasms that are described makes salivary gland tumors one very challenging subject.

Pleomorphic adenoma is a benign salivary gland tumor with variable cytomorphological and architectural manifestations. It is the most common salivary gland tumor [2,10] and the majority is located in the parotid gland [10]. It occurs more frequently in the

third to sixth decade of life, with a female-to-male ratio of 2:1 [10]. They usually present as a slow-growing, unilateral, asymptomatic swelling in the parotid gland [1,10] and multifocal pleomorphic adenomas are very rare [1]. These tumors are composed of a mixture of epithelial and myoepithelial components and the identification of both is necessary for the diagnosis [10]. Parotid pleomorphic adenomas are usually encapsulated, however, frequent recurrence was found following tumor enucleation, which was thought to be due to tumor pseudopodia encroaching beyond the tumor capsule. Changes in the management, regarding wider margins excised beyond the tumor capsule, have reduced recurrence rates to 0-2,5% [1]. Tumor disruption and spillage have also been reported as variables with an independent effect on recurrence [10]. Malignant transformation can occur (3-15%) [1,10] and the risk increases with time, thus, surgical treatment at the time of diagnosis is ideal [1].

The overall most common malignant salivary gland tumor is the mucoepidermoid carcinoma and it is also the most frequent malignancy of the parotid glands (33%) [2] accounting for 4.2-12% of all parotid gland tumors [9]. It occurs over a wide age range with a peak incidence in the second decade of life [10]. Clinical presentation is variable, according to location, size and grade [10]. This tumor is a distinctive salivary gland malignancy composed of mucus-secreting, intermediate (clear-cell), and epidermoid tumor cells in varying proportion [10,11] forming cystic and solid patterns [10]. Based on histological appearance, mucoepidermoid carcinoma is classified into a "high-grade", "intermediate-grade" or "low-grade" malignancy [10]. Low- and intermediate-grade tumors are less aggressive and are generally cured by complete surgical excision [10]. Surgery is the mainstay of treatment for malignant salivary gland tumors and it should be pursued when negative surgical margins can be achieved [11]. The 10-year survival rates for low-, intermediate-, and high-grade mucoepidermoid carcinomas are approximately 90%, 70%, and 25%, respectively [10].

Parotid gland tumors are usually solitary and multiple tumors are uncommon [3,6]. Bilateral parotid gland tumors, either asynchronous or simultaneous are known but rare [9]. These tumors are more frequently benign [6], the most common example being Warthin tumor [9]. Combined benign and malignant lesions, and bilateral tumors of different histologic types are even less frequent [6]. From a comprehensive review of the literature it appears that the occurrence of bilateral primary tumors of benign and malignant histologic types is extremely rare.

In this article, the authors report a rare case of combined benign and malignant tumors of contralateral parotid glands, that presented synchronously. We thoroughly reviewed the literature available on *Pubmed* and could not find another reported case of synchronous bilateral parotid gland tumors with this combination of histologic types, therefore, to the best of our knowledge, this is the first description of a case of synchronous pleomorphic adenoma and mucoepidermoid carcinoma of contralateral parotid glands.

At the present time, the mechanisms involved in the occurrence of bilateral parotid tumors are not clear. The development of multiple Warthin tumors seems to be related to their histological origin [5]. The histogenesis of other types of multiple primary tumors is not clear [5]. Some authors indicated that multiple primary tumors might occur after radiotherapy [5], however, this was not the case in the present patient, since she had no history of radiotherapy. Yu *et al.* stated that the development of multiple primary tumors in the parotid glands, apart from Warthin tumor, may be a coincidence [5].

This case report also illustrates that the preoperative work up of patients that present with a unilateral parotid gland tumor should always include bilateral imaging studies, such as ultrasonography, in order to exclude occult contralateral disease. Our patient presented with a mass on the right parotid gland and did not notably refer the left parotid tumor. Since bilateral parotid tumors are described and can occur, careful preoperative examination is mandatory, including clinical and imagiological examination of the affected and contralateral sides.

Conclusion

Bilateral parotid gland tumors are uncommon, and the histopathology is mostly Warthin tumor. Synchronous bilateral parotid gland tumors with different histologic features are extremely rare, specially combining benign and malignant histologic types.

Although multiple primary parotid tumors are rare, it is important to consider that they can be present. Careful preoperative clinical and imagiological evaluation of bilateral parotid glands is necessary whenever a parotid tumor is identified.

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