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Angaben zur Veröffentlichung / Publication details:

Heine, Daniela, Johannes Zenk, and Georgios Psychogios. 2018. "Two case reports of synchronous unilateral pleomorphic adenoma and cystadenolymphoma of the parotid gland with literature review." *Journal of Ultrasonography* 18 (75): 369–73. https://doi.org/10.15557/jou.2018.0055.

Cite as: Heine D, Zenk J, Psychogios G: Two case reports of synchronous unilateral pleomorphic adenoma and cystadenolymphoma of the parotid gland with literature review. J Ultrason 2018; 18: 369–373.

Submitted: 19.07.2018
Accepted: 31.11.2018
Published: 31.12.2018

Two case reports of synchronous unilateral pleomorphic adenoma and cystadenolymphoma of the parotid gland with literature review

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DOI: 10.15557/JoU.2018.0055

Keywords
synchronous unilateral parotid tumors, pleomorphic adenoma, cystadenolymphoma, B-mode ultrasound, virtual touch imaging quantification (VTIQ)

Abstract

Background: Synchronous unilateral tumors in the parotid glands account for less than 5–10% of all salivary gland neoplasms. Mostly these are cystadenolymphomas, but tumors of different histological types can be found as well. In these cases it is often pleomorphic adenoma in combination with cystadenolymphoma. Ultrasound is the first choice imaging modality. **Case report:** We present two patients with two simultaneous tumors in a unilateral parotid gland. In each case, B-mode ultrasound showed two hypoechoic masses, one of which was predominantly cystic. The subsequent use of Virtual Touch Imaging Quantification (VTIQ) showed tumors of elastic tissue. After a parotidectomy, both cases were diagnosed with pleomorphic adenoma combined with cystadenolymphoma. **Conclusion:** The combination of B-mode ultrasound and VTIQ is an important diagnostic modality and may improve diagnostic accuracy to differentiate between benign and malignant lesions. Every clinician should be aware of the co-existence of different histological types of tumors in unilateral salivary glands.

Salivary gland tumors amount to only 3–6% of all head and neck tumors with most of them occurring in the parotid gland[1]. Pleomorphic adenoma is the most common benign tumor of salivary glands and grows predominantly in the parotid gland. The risk of malignant transformation is about 6%[2,3].

Cystadenolymphomas, also known as Warthin tumors, account for 25–30% of benign tumors in the parotid gland. They present as ipsilateral multicentric in up to 10% of all cases. Patients with cystadenolymphomas are often smokers. Malignant transformation is extremely rare[4].

Synchronous unilateral tumors in the major salivary glands can be found in 5–10% of all salivary gland neoplasms[3]. The majority of these are multifocal cystadenolymphomas[5]. Ipsilateral salivary gland tumors of different histologic types are predominantly pleomorphic adenomas in combination with cystadenolymphomas[6].

Ultrasound is the first choice of diagnostic imaging[1]. Virtual Touch Imaging Quantification (VTIQ) is a new technique that uses acoustic radiation force impulse waves, enabling qualitative and quantitative measurements of the physical stiffness of tissues. Malignant tissue is stiffer than benign tissue (Fig. 1, Fig. 2).

In cases in which the extent of a tumor is difficult to assess in sonography or in cases where tumors grow in the deep lobe of the salivary gland, MRI is of great importance.

The aim of this study was to increase awareness of simultaneously occurring pleomorphic adenomas and cystadenolymphomas and to show that B-mode ultrasound, and

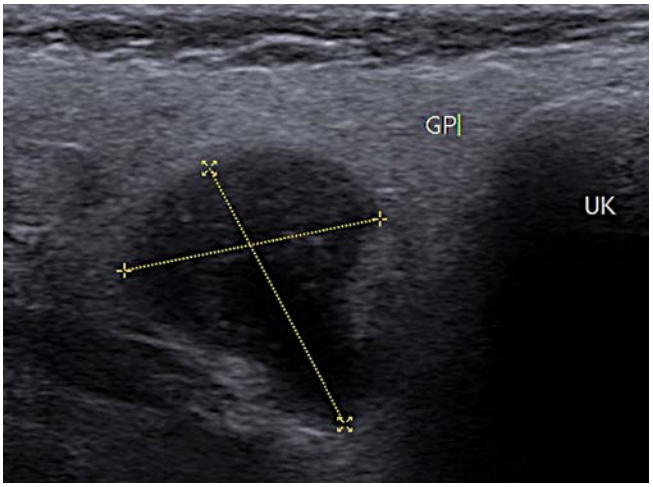

Fig. 1. *B-mode sonography of the first tumor in the caudal pole of the right parotid gland (1. case), GP – parotid gland, UK – mandible*

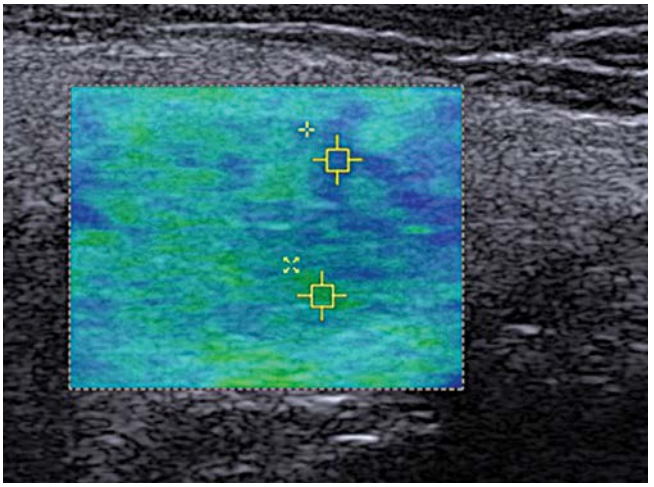

Fig. 2. *Virtual Touch Imaging Quantification of the first tumor in the caudal pole of the right parotid gland (1. case). You can see that the region of interest is measured and shows a colour-graded scale. Blue regions depict tissue that is more elastic than red coloured regions*

elastography (VTIQ) in particular, are important diagnostic modalities in the differential diagnosis of patients.

Case report

In the first case, a 71-year-old female tobacco smoker consulted a physician due to a slowly growing tumor of the right parotid gland she noticed several years earlier. The tumor was palpated preauricularly in the right Regio parotidea. In the physical examination, the facial nerve was intact on both sides.

Sonographically, the first tumor was well-defined, hypoechoic with an anechoic zone and located in the caudal parotid pole. It measured 13.8 × 15.3 × 21.0 mm (Fig. 3). VTIQ showed an elastic tumor (color-rated: blue to green, Fig. 2). This technology utilizes an acoustic push pulse to examine the displacement of tissue elements. This information is used to create a qualitative map. Blue regions depict tissue that is less stiff than red colored regions (Fig. 1, Fig. 2). The second tumor was located over the musculus masseter and measured 32.2 × 15.2 × 31.0 mm. It was well-defined as well, but lobulated. It showed an anechoic area, echogenic internal

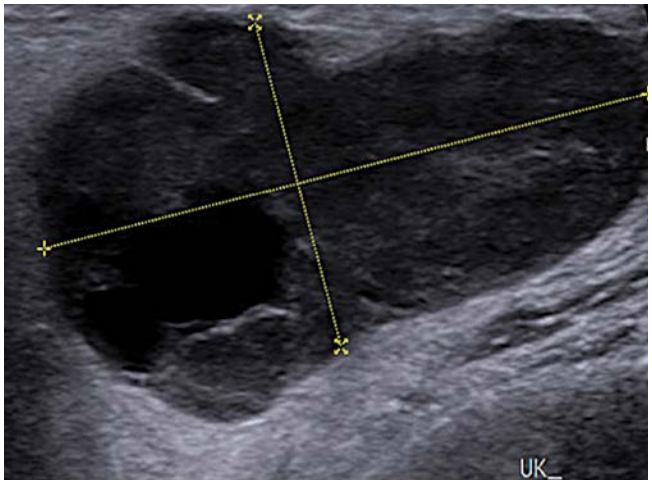

Fig. 3. *B-mode sonography of the second tumor with one cystic area in the right parotid gland (1. case), GP – parotid gland, UK – mandible*

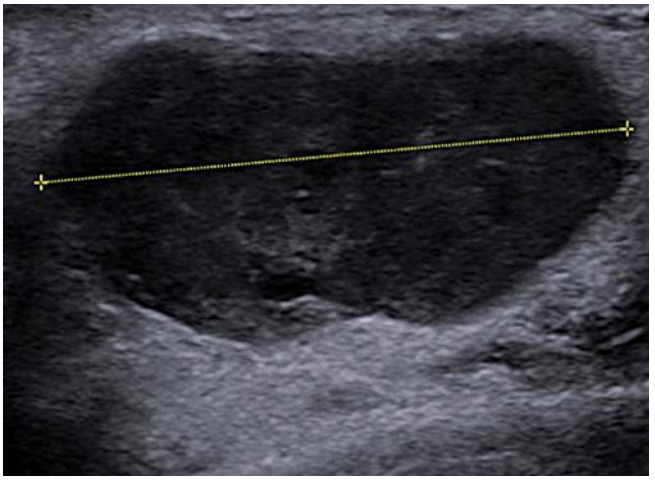

Fig. 4. *B-mode sonography of the second tumor with one cystic area in the right parotid gland (1. case)*

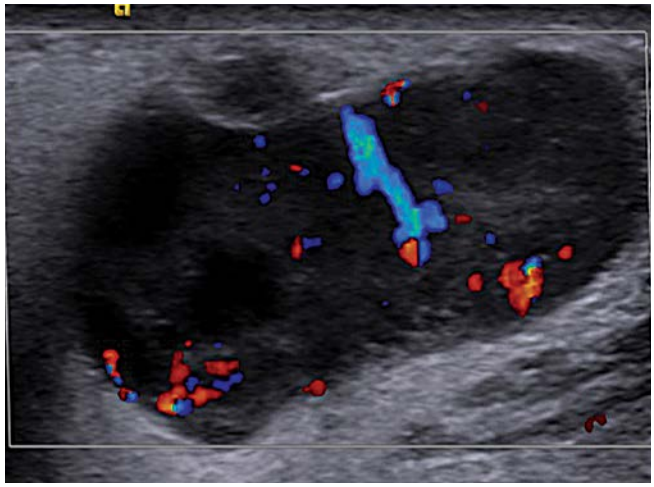

Fig. 5. *Color-coded duplex sonography of the second tumor in the right parotid gland (1. case). It shows a diffusely vascularized pattern with no characteristic pattern and an anechoic area indicating a cystic area*

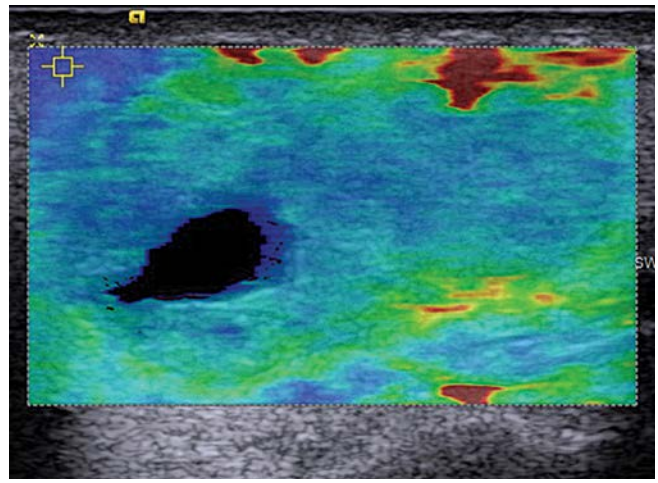

Fig. 6. *Virtual Touch Imaging Quantification of the second tumor with one cystic area ("black area") and a colour-graded scale in the right parotid gland (1. case). Blue regions depict tissue that is more elastic than red coloured regions. The red areas can be artefacts because of the surrounding bone. One measurement shows the velocity of the shear waves*

echoes and distal acoustic enhancement (Fig. 4, Fig. 5). It was diffusely vascularized (Fig. 6). In the VTIQ, the tissue was also less stiff with a "black hole" representing a cystic area (Fig. 2). Due to the different appearance of the two lesions in preoperative ultrasound, the presence of two distinct entities was suspected. Therefore, a decision to perform a complete parotidectomy without prior fine needle aspiration was made. The two lesions were removed and sent for histological examination. The macroscopic differences on the cut surfaces were clearly visible (Fig. 7).

On histopathologic examination, the mass with polycystic areas turned out to be a cystadenolymphoma, whereas the other tumor was diagnosed as a pleomorphic adenoma.

The second case was a 68-year-old woman with two masses in the right parotid gland. On physical examination, two indolent tumors were palpable. Sonographically, two well-defined masses were seen, one preauricular and one in the caudal pole of the right parotid gland. The preauricular, hypoechoic, homogeneous tumor measured 11.5 × 5.6 × 8.2 mm and showed distal acoustic enhancement (Fig. 8). The one in the caudal pole, also hypoechoic and with distal acoustic enhancement, but with echogenic internal echoes, measured 18.9 × 10.2 × 11.8 mm (Fig. 9). Its vascularization was rather peripheral (Fig. 10). Histologically, a pleomorphic adenoma and a cystadenolymphoma were diagnosed as well.

Discussion

Multiple salivary gland tumors account for about 5–10% of parotid gland lesions with Warthin tumors being the most common. As mentioned before, synchronous parotid tumors with different histological types account for less than 1% of all salivary gland tumors and the most common combination is Warthin tumor with pleomorphic adenoma[6]. A few cases of synchronous benign and malignant tumors are reported but they are extremely rare. In these cases, the most common combination is Warthin tumor and mucoepidermoid carcinoma[3].

Preoperative imaging is of great importance. Diagnostic accuracy rates of approximately 90% can be achieved. Sonography is the first choice imaging to

Fig. 7. *Tumor mass after resection (1. case): Both entities together: on the left side beige-browny, lobulated cut surface with a cystic formation (pleomorphic adenoma, arrow), on the right side browny-yellow, polycystic cut surface (cystadenolymphoma, asterisk)*

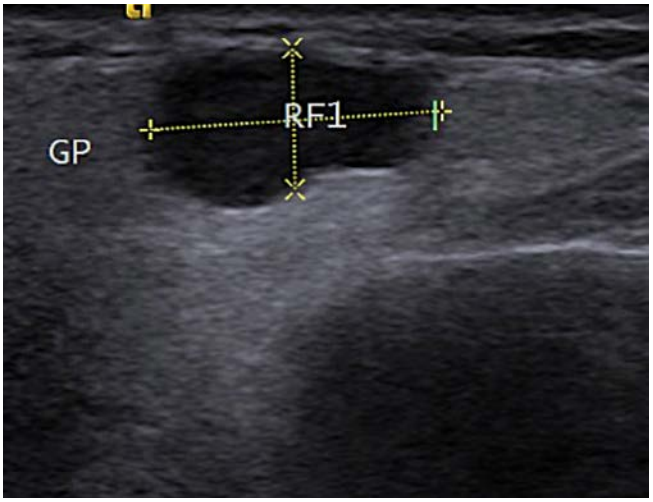

Fig. 8. *B-mode sonography of the first tumor in the preauricular area of the right parotid gland (2. case, RF1 – first tumor)*

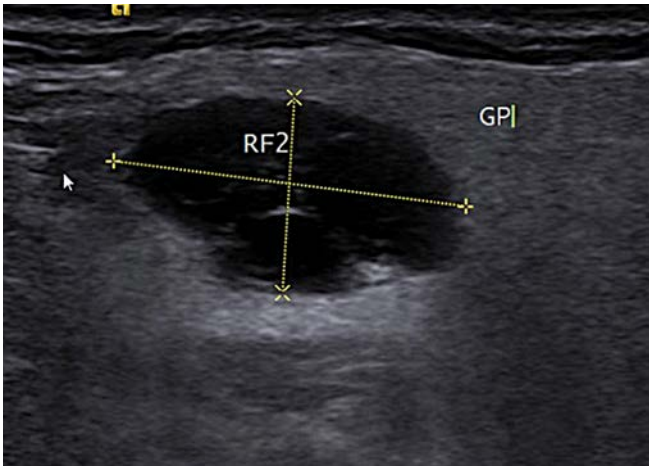

Fig. 9. *B-mode sonography of the second tumor in the caudal pole of the right parotid gland (2. case, RF2 – second tumor)*

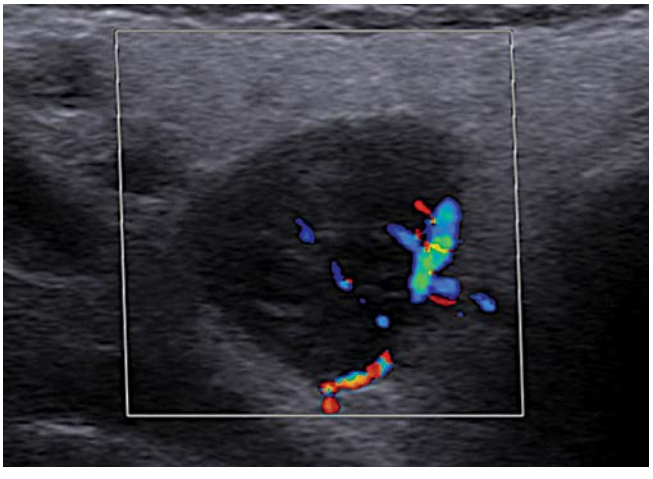

Fig. 10. *Color-coded duplex sonography of one tumor in the right parotid gland which is peripherally vascularized (2. case)*

differentiate between tumor types[1]. Further imaging modalities, predominately MRI, can be used when the tumor is large and sonographically difficult to assess in its extent. Ultrasound and MRI seem to be very efficient in differentiating between benign and malignant lesions[7]. In a study of Mansour *et al.*[8], 202 patients with parotid gland lesions were examined with the use of multimodal ultrasonographic pathway including high-resolution B-mode ultrasound, real-time sonoelastography, color-coded duplex sonography and contrast-enhanced ultrasound.

They concluded that B-mode ultrasound in combination with clinical examination and medical history is able to identify malignant tumors with a sensitivity of up to 77% and with a specificity of up to 98%. The authors suggested that this is a useful method to predict the dignity of tumors, to plan the required surgery and to prevent re-operations[5].

Elastography is a useful additional modality to provide further qualitative and quantitative information about tissue stiffness and to evaluate the dignity[9]. VTIQ is a new technology of two-dimensional shear wave imaging, which provides a combination of quantitative and relative stiffness imaging. In general, malignant tissue is stiff and benign tissue is soft. Pleomorphic adenoma seems to be stiffer than cystadenolymphoma but less stiff than malignant tumors[1].

Preoperative Fine-needle aspiration cytology (FNAC) guided by ultrasound is widely used because it is fast and provides additional information about the tumor's dignity, helping with better treatment planning. Core needle biopsy on the other hand is controversially discussed due to the possible tumor seedling and the risk of fistula formation[1,10].

Complete resection of the tumor with lateral or total parotidectomy is the gold standard[6]. Depending on the dignity of the tumor, an adjuvant therapy may be necessary.

Conclusion

The vast majority of multiple salivary gland tumors are cystadenolymphomas, also known as Warthin's tumors. However, clinicians should be aware of the possible coexistence of tumors of different histological types in ipsilateral salivary glands. B-mode ultrasound in combination with color-coded duplex sonography and VTIQ may improve the accuracy of preoperative diagnosis. Ultrasound is the most important modality to predict the dignity and to plan required surgery.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organizations, which might negatively affect the contents of his publication and/or claim authorship rights to this publication.

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