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Parotid gland oncocytoma mimicking local malignancy of Warthin's tumour on contrast-enhanced ultrasound

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Parotid gland oncocytoma (PGO) is a rare benign epithelial tumour that usually occurs in the female population; it is also called oncocytic adenoma, oxyphilic granular cell adenoma, or oxyphilic adenoma [1]. PGO is considered rare, representing less than 1% of parotid glands, and the risk factors of PGO are as yet unknown. Its symptoms may be non-specific, such as pain, discomfort, or facial nerve paralysis. Contrast-enhanced ultrasound (CEUS) plays an important role in the diagnosis of parotid lesions such as PGO, given its noninvasiveness, low cost, widely availability, and lack of ionizing radiation. Because of its low prevalence and overlap with other parotid tumours, the accurate diagnosis of PGO remains a challenge. We present the following article in accordance with the CARE reporting checklist (available at <http://dx.doi.org/10.21037/apm-20-972>).

There are no previous reports of the performance of PGO mimicking local malignancy of Warthin's tumour (WT) on CEUS. Herein, we describe a case of PGO mimicking other parotid lesions, which was identified postoperatively in a 57-year-old female.

A 57-year-old woman who presented with a 24-month history of an enlarging mass over the left parotid gland underwent ultrasonography at her local hospital, which revealed a mass in the left parotid. She was systemically well with no weight loss, facial pain, dysphagia, xerostomia, or facial weakness. She had no family history of parotid cancer. On physical examination, there was a medium to hard, smooth, ill-defined, painless mass in the left parotid region. The skin overlying the surface of the mass was normal. There was no cervical and submandibular lymphadenopathy. The facial nerve was not involved. There were no aberrant haemato-biochemical findings. CEUS comprised injecting a 4.8 mL bolus of

Sonovue (Bracco, Milan, Italy) intravenously followed by a 5 mL saline flush. The overall lesion showed heterogeneous and centripetal hyper-enhancement. A partial heterogeneous lesion was located in the anterior edge of the nodule, which was previously washed out. The remaining part showed sustained hyper-enhancement. The CEUS revealed local malignancy of WT (Fig. 1). For further characterization of the mass, contrast-enhanced computed tomography (CECT) of the parotid gland was performed, which showed a lobulated mass in the left parotid gland measuring approximately 2.1 × 1.8 × 2.7 cm without infiltrating the surrounding tissue. CECT revealed possible pleomorphic adenoma (Fig. 1). Core needle biopsy (CNB) performed prior to our evaluation revealed an abundance of eosinophilic cells in the cytoplasm, tending toward eosinophilic tumour. Histology demonstrated PGO (Fig. 1). The patient received partial excision of the left parotid gland and preserving the facial nerve. There were no long-term postoperative complications, and the patient is disease-free at 2 years.

PGO is a rare benign tumour that presents a diagnostic challenge to clinicians. The tumour can also arise in other organs, such as the kidney, adrenal, pituitary glands, and thyroid. PGO can be multifocal or bilateral in the parotid gland, which typically affects individuals between the sixth and eighth decades of life. The real cause of PGO is unknown, but it may be associated with 5 or more years' history of radiation exposure. PGO typically present as a painless and mobile mass with clinical complaint of swelling for weeks to years. WT is a common and benign tumour, which consists of epithelial and lymphoid tissue. The epithelial or lymphoid component of WT has a rare possibility of malignant



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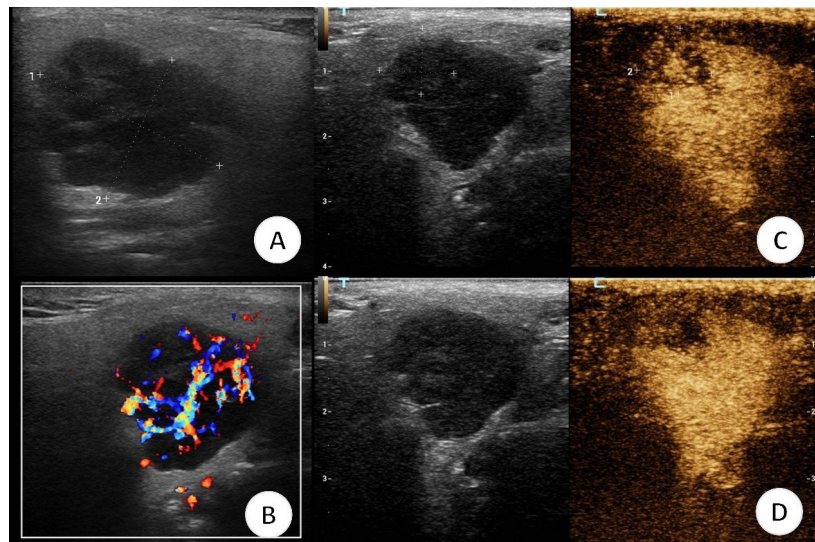


Figure 1. Contrast enhanced ultrasound (CEUS) images at 11 s (A) and at 48 s (B) from the SonoVue injection. Partial heterogeneous enhancement was located in the anterior edge of the lesion, which was previously washed out. Contrast-enhanced computed tomography (CECT) of the parotid gland showing that the mass was rapidly enhanced in the early arterial phase (C) and obviously highly enhanced in the late arterial phase (D). There were no obvious enlarged lymph nodes on either side of the neck, and the right parotid gland was normal. Histopathological slide (E) demonstrating a large mass with a fibrous envelope surrounded by normal parotid gland tissue (haematoxylin and eosin staining, $\times 4.8$). Pathological sample showed abundant oncocytes (F) of the mass (haematoxylin and eosin staining, $\times 100$). Immunohistochemically, the result of the test using EMA is positive ($\times 200$) (G). Ki-67 expression $< 5\%$ ($\times 200$) (H)

transformation. PGO have cells with centrally located pycnotic nucleus and pleomorphic mitochondria in their cytoplasm, and they are big cell tumours characterized by acinar and ductal cell metaplasia.

Histopathologically it is a diagnostic challenge for the differentiation of PGO and WT, because both comprise cell types rich in mitochondria [2]. PGO is described as well-defined masses with heterogeneous enhancement on ultrasound, which may lead to misdiagnosis as WT. PGO demonstrates increased radioactivity absorbance and increased extraction with delayed extraction function on PET imaging. The US and CEUS features of PGO are nonspecific and complex. The echo of the tumour can be inhomogeneous if the lesion has cystic or haemorrhagic areas. The classic central scar and spoke-wheel vascular pattern is not often observed. Colour Doppler gives information of the vascular architecture and vascular distribution of a lesion. CEUS provides information on the perfusion pattern of a lesion and perfusion down to the capillary level [3].

Pleomorphic adenoma and WT are the most commonly diagnosed histological types of benign parotid lesions [4]. Pleomorphic adenoma can lead to recurrence or malignancy, while WT is less prone to malignancy or recurrence. In this case, a partial heterogeneous lesion was located in the anterior edge of the nodule, which was previously washed out. The previous wash out was mistaken as the malignant part of the mass. The variability

of radiological findings and pleomorphic histological appearance highlights the diagnostic challenges of PGO.

PGO is a rare disease that should be considered in the differential diagnosis of parotid lesions. For patients with an indefinite diagnosis or symptoms, a complete resection is recommended.

Authorship

P.H. designed this study. J.L. and E.H. acquired the data. J.L. wrote the main manuscript text. All authors reviewed the manuscript. The authors confirm no conflict of interests related to this study.

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