



Case Report

Bilateral parotid oncocytoma with spontaneous intratumoral hemorrhage: a rare hypervascular parotid tumor with ASL perfusion



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ABSTRACT

We present a rare case of bilateral parotid oncocytoma with spontaneous intratumoral hemorrhage. Magnetic resonance imaging revealed multiple, bilateral, well-defined masses within the parotid glands with increased vascularity based on arterial spin labeling Magnetic Resonance (MR) perfusion imaging and restricted diffusion on diffusion-weighted imaging. During the clinical follow-up, the patient developed extensive hemorrhage from the largest tumor, presumably due to a combination of the hypervascular nature of oncocytoma and hemodialysis.

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1. Introduction

An oncocytoma is a rare salivary gland tumor and consists of about 1% of all salivary gland tumors [1] and 0.5 to 1.2% of all parotid tumors [2]. Only 7% of oncocytomas have been reported to be bilateral [1]. We describe an unusual case of bilateral parotid oncocytoma that later developed spontaneous intratumoral hemorrhage with computed tomography (CT) and MRI findings.

2. Case report

An 82-year-old woman was referred for evaluation of her left parotid mass, which was incidentally noticed by a physician. The mass was firm without tenderness to palpation. She was on hemodialysis due to chronic renal failure and had no history of smoking or other significant medical history.

Imaging examination revealed two large left parotid masses (37 × 18 × 46 mm and 35 × 21 × 38 mm), one in the deep lobe extending to the parapharyngeal space and another in the superficial lobe. There was a single large mass (21 × 17 × 34 mm) in the superficial

lobe of the right parotid gland. All parotid lesions were well circumscribed and had a smooth and partially lobulated contour without calcification.

The masses demonstrated homogeneously high attenuation on noncontrast CT, and CT Hounsfield values of the left deep lobe mass were 60 HU (Fig. 1A). On MRI, the lesions appeared homogeneously hypointense to parotid parenchyma on T1-weighted images (T1WI) and isointense on fat-saturated T2-weighted images (T2WI) and short-tau inversion-recovery (STIR) images (Fig. 1B–D). The left deep lobe mass demonstrated multiple flow voids, corresponding to neovascularity on noncontrast three-dimensional time-of-flight MR angiography (TOF-MRA) (Fig. 2A). All lesions also showed high signal intensity on arterial spin labeling perfusion image (ASL-PWI) (2D pulsed echo-planar imaging, TR 2500 ms, TE 12 ms, TI1 700 ms, TI2 1800 ms) (Fig. 2B), suggesting a hypervascular nature of these masses. These masses were associated with restricted diffusion with apparent diffusion coefficient (ADC) values of $0.8\text{--}1.0 \times 10^{-3} \text{ mm}^2/\text{s}$ [diffusion weighted image (DWI): single-shot echo-planar imaging] (Fig. 2C, D). No imaging findings to suggest perineural tumor extension along the facial nerves were present. Contrast materials were not administered to the patient, due to concern for nephrogenic systemic fibrosis.

A fine needle aspiration cytology for the left deep parotid mass revealed clusters of proliferation of oncocytic cells showing bland nuclear features, consistent with oncocytoma. Due to the benign nature of the tumor, advanced age, and patient's medical comorbidity, the patient was placed under clinical observation and imaging follow-up. There had been no change in size or morphology for 2 1/2 years, and then,

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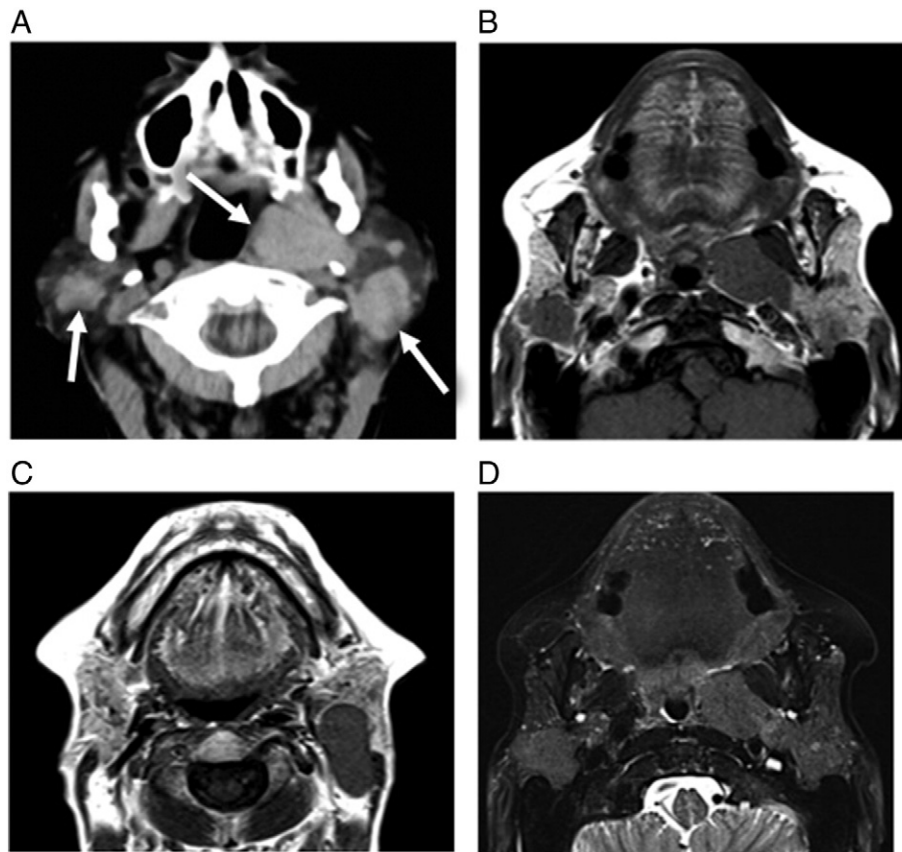


Fig. 1. (A) Noncontrast CT (axial image), (B, C) noncontrast T1WI (axial image), (D) fat-saturated T2WI (axial image): high-density parotid masses are seen in bilateral superficial lobe and right deep lobe (arrows, A). The masses show hypointense signal on noncontrast T1WI and isointense signal on fat-saturated T2WI (B–D).

she presented with acute onset of her left facial pain and swelling without a history of recent trauma.

MRI revealed marked interval increase in the size of the left superficial lobe tumor. The signal intensity had also changed to mild hyperintense signal on T1WI and marked hyperintense signal on T2WI compared to the initial MRI images, suggesting intratumoral subacute hemorrhage (Fig. 3). In addition, there was interval development of periglandular edema and inflammation. The other two parotid tumors showed no changes in imaging appearance. Spontaneous intratumoral hemorrhage was likely caused by a combination of hypervascular nature of oncocytoma and coagulopathy related to hemodialysis.

3. Discussion

Oncocytomas represent about 1% of the salivary gland neoplasms, and 84% of them occur in the parotid gland [1]. Oncocytomas most commonly present in the 6–8th decades as a painless facial mass. No differences regarding gender predilection was reported [1], although some reported slightly higher frequency in females [3]. Bilateral multiple oncocytomas are seen in 7% of cases, often distributing in both the superficial and deep lobes [1,4].

Oncocytomas are often present with well-defined borders with the smooth or lobulated contour on CT and MRI [4,5]. They most commonly demonstrate hypointense signal on both T1WI and T2WI compared to the normal parotid gland [1,5–8], isointense signal on STIR images, fat-saturated T2WI, and fat-saturated postcontrast T1WI [5,6].

An oncocytoma is a histologically hypercellular tumor and consists of solid clusters or cords of tightly packed oncocytes separated by thin strands of fibrovascular stroma [3], which is suggested by the low signal

intensity on T2WI and low ADC values [8–10]. Kasai et al. reported that ADC values of oncocytoma ranged from 1.0 to $1.16 \times 10^{-3} \text{ mm}^2/\text{s}$ [6]. The high density on noncontrast CT is most likely due to hypercellularity of the oncocytoma in our case. Oncocytomas commonly show early enhancement and early washout on dynamic contrast-enhanced MR images (dynamic MRI) [6], supporting increased tumor vascularity [11]. Kato et al. recently reported that Warthin tumors show significant hyperintense signal on ASL-PWI compared to a malignant parotid tumor and pleomorphic adenoma [12]. Hyperintense signal on ASL-PWI as well as abundant neovascularity on TOF-MRA in the present case, therefore, suggests increased vascularity, while the relatively small amount of extravascular extracellular space may explain the early washout on dynamic MRI [5].

Oncocytomas might be indistinguishable from Warthin tumors by traditional cross-sectional imaging alone. Warthin tumor is the second most common parotid tumor [1] and the most common bilateral parotid tumor [13]. It presents bilaterally in 5–14% and can be multicentric in 12–20% of cases [1] and occurs in the parotid tail in 87% of cases [14]. It is composed of both cystic and solid components, which consist of epithelial cells and lymphoid stroma with fibrovascular tissue associated with high cellularity and hypervascularity [1]. Therefore, the solid component demonstrates hypointense signal intensity on T2WI with low ADC values and early enhancement with early washout of the tumors on dynamic MRI [12,15]. Warthin tumors, however, have a strong association with cigarettes smoking [16] and male gender predilection, as well as the presence of cystic components, differentiating it from an oncocytoma.

The other differential possibilities of multiple bilateral parotid malignant tumors include acinic cell carcinoma, malignant lymphoma, and metastasis. Bilateral acinic cell carcinoma occurs in 3–5% of cases [17] and has been reported to show isointense to hyperintense signal on

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