CASE REPORT

A case of cystadenocarcinoma of the ectopic salivary gland: comparison of pre-operative ultrasound, CT and MR images with the pathological specimen

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Cystadenocarcinoma is a rare salivary gland tumour. Only a few case studies have provided pre-operative images of these tumours. This report demonstrates the case of a 28-year-old male with cystadenocarcinoma arising from an ectopic salivary gland with lymph node metastasis in the right upper neck. Ultrasound including Doppler images showed two masses with scant vascular flow. One was a hyperechoic mass enclosed within a low echoic cystic lesion and the other was a solid hypoechoic mass. Contrast enhancement CT scans demonstrated a ring enhanced mass and weakly homogeneous enhanced masses in the right upper neck. Dynamic studies showed increased enhancement in delayed phase CT that was the same as that in other malignant salivary gland tumours. Moderate to slightly high signal intensity was seen on T_1 weighted MR images and axial T_2 weighted MR images showed one heterogeneous mass in a high signal lesion and a moderate to high signal intensity mass. The authors discuss the pre-operative findings of ultrasound with Doppler imaging of this neoplasm, and CT findings including dynamic study images and MRI, comparing the findings with the post-operative pathological features of the tumour.

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Introduction

Cystadenocarcinoma is a rare malignant tumour of the salivary glands described by the World Health Organization in 1991.¹ The estimated incidence is about 2–5% of malignant salivary gland tumours, and lymph node metastasis rarely occurs.^{1–3} Pre-operative ultrasound, CT and MRI for pleomorphic adenoma and monomorphic adenoma of the salivary gland are well documented and have provided diagnostic utility in previous reports.^{4–6} However, only a few reports have shown the pre-operative imaging for cystadenocarcinoma of the salivary gland owing to the low incidence of the disease.^{7–10} We herein report a case of cystadenocarcinoma with

ipsilateral lymph node metastasis derived from the right upper neck and compare the pre-operative imaging findings, including those from ultrasound, CT and MRI, with the pathological findings.

Case report

A 28-year-old male patient was admitted to Noguchi Thyroid Clinic with a right neck mass that was first noted 6 months previously. The patient did not complain of any other prominent symptoms, such as facial nerve palsy, and had an unremarkable history. Palpation of the left upper neck revealed a 50 mm diameter well-circumscribed elliptical mass.

Ultrasound, including Doppler images using a Philips/ATL HDI 5000 system with a 5 MHz to 12 MHz linear

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Figure 1 Right cervical transverse ultrasound showing two masses (a, c). The primary site is demonstrated as a hyperechoic mass within the low echoic cystic lesion and metastatic lymph node demonstrated as a hypoechoic mass. Doppler sonography of these masses showed scant vascular flow (b, d)

array transducer (Philips Medical Systems, Bothell, WA), showed two oval masses with scant vascular flow (Figure 1). One was a hyperechoic mass enclosed within a low echoic cystic lesion and the other was a hypoechoic mass. Neck CT scanning was performed to assess vascular invasion and the location of the tumour, as well as for differential diagnosis. The enhanced dynamic scans were acquired after the unenhanced scans (Figure 2). The dynamic scans were obtained using a GE High Speed NXi (dual slice) CT scanner (GE Healthcare, Tokyo, Japan). A 100 ml intravenous bolus of a non-ionic iodinated contrast medium (iohexol) was administered *via* the antecubital vein at a rate of 2 ml s^{-1} using an auto-injector. Three series of CT scans were

obtained at 30 s, 90 s and 150 s after injection of the contrast medium. The CT scanning parameters were section thickness of 3 mm, 120 kV, 136–293 mA and a 512×512 matrix. CT showed a ring enhanced mass and weakly enhanced masses in the right upper neck. The lower tumour was attached but had not invaded the internal jugular vein. The peak enhancement at 90 s was observed by a dynamic study (Figure 2k).

MRI was performed using a 0.5 T MRI unit (FLEX ART, Toshiba Medical Systems, Tokyo, Japan). The repetition time/echo time was 75 ms/15 ms for T_1 weighted spin-echo and 3400 ms/102 ms for T_2 weighted fast spin-echo. A 20 cm field of view, 5 mm thick sections at 6 mm intervals and a 256×256 matrix were used. There were

Figure 2 A contrast enhanced CT scan of the primary site shows a bull's eye with ring enhancement in the right upper neck (a–e). The enlarged lymph node was shown as a diffuse enhanced round mass (f–j). The dynamic study showed increased enhancement in the later phase CT of both the Primary tumour site (a–c) and the metastatic lymph node (f–h). Reconstructed coronal and sagittal CT images showed the relationship between tumour sites and the parotid gland (d, e, i, j). The time–CT number (Hounsfield unit) curve of tumour sites showed rapid contrast enhancement at 30 s and peak enhancement at 90 s (k)

two moderate-to-slightly high signal intensity masses on T_1 weighted images (Figure 3). Axial T_2 weighted MR images showed one heterogeneous mass in a high signal lesion and one moderate-to-high signal intensity mass.

The fine-needle aspiration cytology (FNAC) showed a large number of oval tumour cells with or without

vacuoles which were suggestive of mucoepidermoid carcinoma. The pre-operative findings confirmed the parotid carcinoma with neck lymph node metastasis and surgical resection was planned. *En bloc* surgical resection with lymph node dissection was performed under general anaesthesia. No surgical complications

Figure 3 MR images of cystadenocarcinoma. Axial T_1 weighted images demonstrated both of the two slightly high signal intensity masses were smooth (a, arrow; c, arrow). Axial T_2 weighted MR images showed a heterogeneous mass enclosed within a hyperintense lesion for the primary tumour (b, arrow) and intermediate-to-high signal intensity for the metastatic lymph node (d, arrow)

developed post-operatively. A final diagnosis of cystadenocarcinoma was made based on the pathological features, as shown in Figure 4. There has been no evidence of either recurrence or metastasis after 18 months of follow up.

Discussion

Cystadenocarcinoma is uncommon and a large majority of the reports on the disease have been case presentations. The most common locations of this

cancer are the parotid, sublingual and minor salivary glands such as the lip or buccal, while occurrence in other areas is extremely rare.^{3,9–13} The present case was a rare case of cystadenocarcinoma derived from an ectopic salivary gland, confirmed by pathological and intraoperative findings, in which the tumour was detached from parotid and submandibular glands.

Cystadenocarcinoma grows slowly and rarely presents with lymph node metastasis or distant metastasis.^{2,3} In the presented case, the ipsilateral lymph node metastases were confirmed post-operatively. We diagnosed the metastatic lymph node based on the existence of the lymphatic sinus.

Figure 4 Gross findings (a) and histological features of the tumour (b–d). The cut surface of the tumour showed the solid mass inside a cystic lesion (a, arrow) and homogeneous masses in the right upper neck (a, arrowhead). The cystic lumen exhibits various degrees of papillary formation (b, c). The metastatic lymph node had the lymphatic sinus (d, arrow)

The primary lesion was found to be devoid of lymphatic structures in the pathological specimen. Post-operative pathology confirmed that the metastatic lymph node was attached to the lower edge of the right parotid gland (Figure 2i). The primary lesion was located under the metastatic lymph node and far from the parotid and the submandibular glands. We diagnosed that this case was a cystadenocarcinoma derived from the ectopic salivary gland situated below the metastatic lymph node.

The pre-operative imaging of the primary tumour shown in Figures 1a,b, 2a–e and 3a,b are consistent

with the solid mass inside a cystic lesion. These findings agree with gross histopathological features (Figure 4a). Koç *et al*⁷ previously presented the MRI findings of a patient with cystadenocarcinoma of the submandibular gland. They demonstrated a hypointense, non-homogeneous smooth mass on axial T_1 weighted images and heterogeneous hyperintensity composed of solid and cystic lesions on axial T_2 weighted images. This is the first report of a cystadenocarcinoma that compared pre-operative ultrasound, CT and MRI findings with the pathological specimen.

The neoplasms originating from the salivary gland have many histological types, ranging from benign to high-grade malignancy. If the neoplasm is a low-grade malignancy, CT scans or MRI may not always reveal typical findings such as an infiltrating mass. Therefore, FNAC is usually performed for differential diagnoses. However, it may be difficult to distinguish the subtype of malignancy pre-operatively. The diagnostic value of pre-operative MRI and FNAC was considered to be a maximum of 80% in terms of sensitivity and specificity.^{14,15} In fact, the FNAC performed for our case diagnosed the patients pre-operatively with a mucoepidermoid carcinoma.

It is difficult to distinguish cystadenocarcinoma from other salivary gland tumours by pre-operative findings including FNAC. However, adenoid cystic carcinoma (ACC), which is one of the more common salivary gland carcinomas, may be distinguished by CT and/or MRI findings. ACC usually has an enhanced and well-circumscribed solid mass without cystic formation.

Contrast enhanced CT scan and MRI imaging of the metastatic lymph node (Figures 2f–j and 3c,d) was similar to the findings of the solid component of the primary tumour. These findings may reflect the homogeneous solid components on gross sections of the

specimen (Figure 4a). The ultrasound for the metastatic lymph node (Figure 1c) showed a hypoechoic mass with slightly heterogeneous echotexture, which had different features compared with the solid component of the primary site. Acoustic enhancement may have contributed to the difference in echoic intensity.

The dynamic CT findings are very important for identifying benign and malignant parotid tumours.^{16–18} Benign salivary gland tumours such as Warthin tumour and basal cell adenoma show intense enhancement in the early phase and decrease gradually in the later phase.^{18,19} In contrast, pleomorphic adenoma shows increased enhancement in the delayed phase CT and malignant salivary gland tumours (such as adenocarcinoma) show peak enhancement at 90 s. The present case showed the malignant salivary gland tumour pattern. To the best of our knowledge, this is the first reported case that describes the dynamic CT findings of cystadenocarcinoma.

In conclusion, we herein reported the pre-operative imaging and pathological findings of cystadenocarcinoma. Cystadenocarcinoma should be included in differential diagnosis if an upper neck mass including the salivary gland showing a cystic component in pre-operative imaging.

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