

Cystic carcinoma of the neck

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ABSTRACT

Malignant cystic lesions of the lateral side of the neck are usually due to metastasis and are rarely primary carcinoma. The most common is metastatic cystic squamous cell carcinoma arising from a primary site at the Waldeyer's ring. However, they can arise from papillary carcinoma of thyroid, lung and mediastinum but very rarely from the abdominal and pelvic organs without regional lymph node involvement. Therefore, accurate diagnosis is very important for appropriate management. We report two cases of cystic squamous cell carcinoma of the lateral neck; cystic metastasis from an occult squamous cell carcinoma of the tongue and a primary branchiogenic carcinoma. We report two cases of cystic carcinoma of the neck of different aetiologies, a cystic metastatic tongue squamous cell carcinoma where the primary was detected on random biopsies during panendoscopy and the other a primary branchiogenic carcinoma. We emphasize the importance of meticulous search for the primary malignant lesions in cases of cystic carcinoma of the neck.

Keywords: Branchial cyst, carcinoma, cysts, metastasis

INTRODUCTION

Cystic neck lesions can cause diagnostic dilemma and require thorough evaluation as they can be neoplastic. Most of the lateral cystic lesions of the neck are benign. Metastatic lesions in the neck are usually solid and rarely cystic. Malignant transformation of a branchial cyst is extremely rare.¹ However controversies remain regarding the existence of branchial cyst or branchiogenic carcinoma.

ment with a right cystic neck lump of six months duration. There were no other palpable lesions in the neck. There was no history of epistaxis, dysphagia or loss of hearing. Blood investigations, Epstein Barr virus status and chest radiography were normal. Magnetic resonance imaging (MRI) of the head and neck region showed no abnormality other than the cystic lesion.

CASE REPORT

Case 1: A 43-year-old Caucasian lady presented to the Otorhinolaryngology depart-

Fine needle aspirate of the cyst revealed anucleated squames and keratin fragments suggestive of a branchial cyst. Excision biopsy was carried out and this showed a cystic lesion with irregularly thickened wall (5 x 3 cm). Microscopy revealed several layers thick squamous cells in the cyst wall which consisted of mixture of non-keratinizing and ker-

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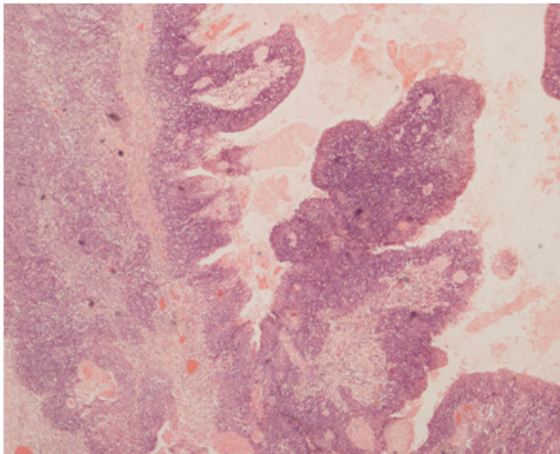


Fig 1: Lymph node, cystic squamous cell carcinoma metastasis from the tongue (H&E stain, x10).

keratinising squamous cells with ill-defined epithelial perls. There was marked nuclear pleomorphism and high mitotic index (Fig. 1). A moderate lymphocytic infiltrate was seen surrounding the malignant epithelial cells. A few subcapsular sinuses were seen suggestive of lymph node origin of the cyst. A diagnosis of metastatic squamous cell carcinoma was made and the principle doctors were advised to evaluate further to rule out the possibility of a primary originating from the Waldeyer's ring.

Panendoscopy with random biopsies of the hypopharynx, base of the tongue, right

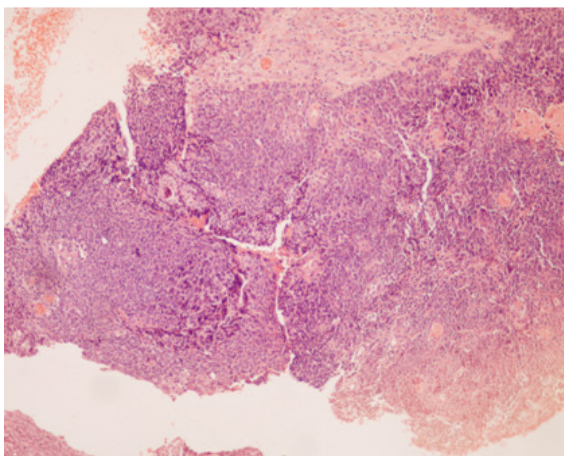


Fig 2: Tongue, primary squamous cell carcinoma (H&E stain, x10).

nasopharynx and the right tonsil were carried out. Biopsies of the base of the tongue was positive for a moderately differentiated squamous cell carcinoma (Fig. 2) which was the primary. She left the country for radiotherapy and was lost to follow-up.

Case 2: A 42-year-old Malay lady presented to the Head and Neck department of RIPAS Hospital with a three-month history of a rapidly growing cystic lesion on the left side of the neck. Blood investigations and chest radiography were normal. A MRI scan of the neck showed a possible branchial cyst with an irregular cyst wall. The cyst (8 x 4 x 1 cm) was excised along with adjacent fibro-fatty tissue that contained three lymph nodes (largest 1 x 0.7 cm). Histology showed the cyst to be lined by squamous epithelium of varying thickness. The squamous cells showed malignant features like hyperchromatic, pleomorphic nuclei with well defined nucleoli and mitotic figures (Fig. 3). There were also foci of non-malignant squamous epithelium with transformation into malignant squamous epithelium (Fig. 4). A marked lymphocytic infiltrate with follicle formation was seen in the cyst wall. The tumour was confined to the cyst wall. The resected lymph nodes showed only reactive changes.

Panendoscopy with random biopsies of the nasopharynx, hypopharynx, base of the tongue and bilateral tonsillectomy were negative for malignancy. The final diagnosis was primary branchiogenic carcinoma. The patient underwent radiotherapy and is well four and half years after the diagnosis. Regular six monthly panendoscopy had so far been negative.

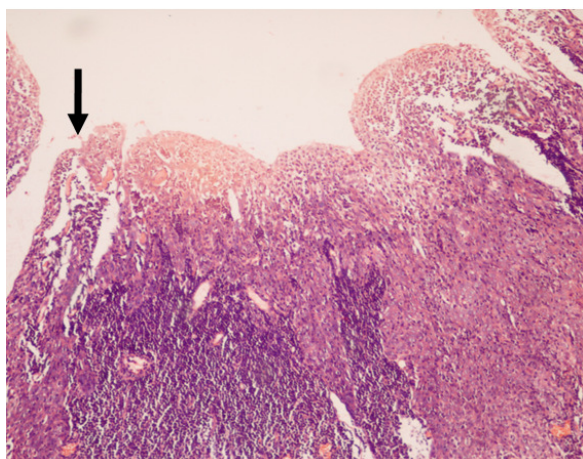


Fig 3: Branchiogenic carcinoma with transformation (an essential feature) from benign to malignant epithelium (black arrow) (H&E, x10).

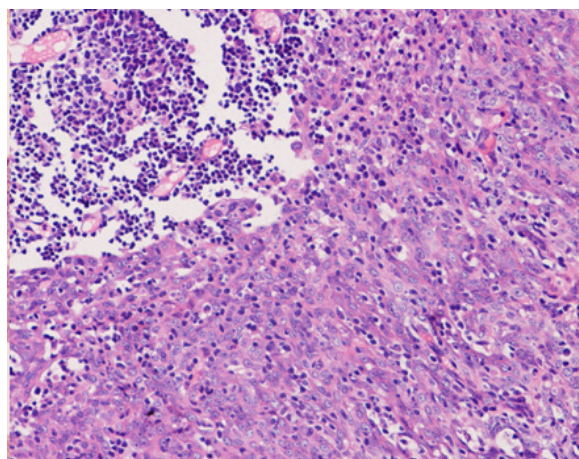


Fig 4: High power view of the branchiogenic carcinoma (H&E, x20).

DISCUSSION

Masses of the lateral neck may be cystic or solid and benign or malignant. Solid masses are always malignant with the exception of a few inflammatory conditions such as tuberculosis. Malignant nodes are usually metastasis from the head and neck region, including the thyroid gland. Metastases from intra-thoracic are rare and even rarer from intra-abdominal organs without other lymph nodes involved. All lesions should be biopsied. Tuberculosis should always be excluded if the lesion is inflammatory especially in countries where tuberculosis remains common. Malignant lesions are either epithelial origin or malignant lymphomas. Epithelial metastases are usually adenocarcinoma, papillary carcinoma or squamous cell carcinomas. Metastatic adenocarcinoma is usually from the lung or upper gastrointestinal tract. Papillary carcinoma is usually from the thyroid but can also be from the lung. Squamous cell carcinoma metastasis is usually from the head and neck region. Malignant lymphomas can either be Hodgkin's or non-Hodgkin's type.

In contrast, cystic lesions of the lat-

eral side of the neck are usually benign. These are branchial cysts. Cystic lesions lined with malignant epithelium are commonly metastasis from occult malignancy in the head and neck region. The other possibility is malignant change in the branchial cyst resulting in the branchiogenic carcinoma as in our second case. However, controversies remain regarding the existence of this entity.

Von Volkmann reported the first case of branchiogenic carcinoma in 1881.⁵ He postulated that malignant lesions in the lateral side of the neck arise from the branchial cleft cysts. The largest series of branchiogenic carcinoma consisting of 28 cases was described by Crile and Kerns in 1935.⁶ Martin *et al* in 1950 proposed criteria for diagnosis of branchiogenic carcinoma (Table 1).⁷

The first criterion assumed that lesions anterior to the tragus are branchiogenic carcinoma. However, metastases especially from the head and neck primaries can occur in this region. Therefore, location alone is unreliable to diagnose branchiogenic carcinoma.

Table 1: Criteria for diagnosis of lateral cystic by Martin *et al.*

1	The cervical tumour must occur anterior to the tragus along the anterior border of the sternomastoid muscle up to the clavicle.
2	The histological appearance supports origin from tissue known to be arising from the bronchial cleft derived structure.
3	Histology supports the carcinoma arising from the wall of the epithelial lined cyst in the lateral aspect of the neck.
4	A minimum five years of follow up which demonstrates no evidence of primary tumour in the head and neck region.

The second criterion taken alone may not also be helpful. The branchial cleft derived structures are lined by squamous cells. Hence complete malignant transformation in a branchial cyst cannot be differentiated from a cystic metastatic squamous cell carcinoma. The third criterion is more supportive of existence of branchiogenic carcinoma. The presence of a transition zone from normal mucosa to malignant mucosa suggests malignant transformation of branchial cyst. The final criterion assumed that the occult primaries would become evident within five years of detecting the metastasis. However, treatment for branchiogenic carcinoma includes radiotherapy and as such the primary if present may no longer be evident later. Furthermore, there are reports that the primary carcinoma have developed later than five years.⁸

Importantly, there are now more so-

phisticated investigations such as high resolution computed tomography (CT) scan, MRI and positron emission tomogram (PET) scan available for detecting primary sites. Therefore, these investigations should be included in the investigating protocol of possible branchiogenic carcinoma.

There are useful clue to the aetiologies of cystic lesions. The age of the patient and duration of the mass is very important. About ten percent of patients above the age of 40 with lateral neck cysts have evidence of malignancy after thorough investigations. Unlike branchiogenic malignancies, cystic metastases tend to appear suddenly and enlarge rapidly. Despite being reported in the literatures, controversies remain regarding the existence of branchiogenic carcinoma, as some believe that with thorough investigations, the primaries can be detected, usually within the

Table 2: A proposed investigation protocol for cystic neck lumps.

1	Detailed history including duration of the cyst and risk factors for head and neck carcinoma (smoking and alcohol consumption).
2	Biopsy for the diagnosis of malignant change
3	CT, MRI and PET scan for detection of the hot spots of the primary lesion
4	Excision of the cyst with frozen section diagnosis if necessary
5	Panendoscopy with random biopsies and bilateral tonsillectomy for detection of the primary site

head and neck region.²⁻⁴ We believe that branchiogenic carcinoma exists, however it remains a rare entity. We also believe that Martin's criteria complemented by a few other investigative techniques, it is possible to differentiate between primary branchiogenic carcinoma and cystic metastasis. We propose an investigation protocol for the evaluation of neck cystic swelling (Table 2) that include the use of some of the more advanced imaging such as MRI and PET scan.

In conclusion, our two cases highlighted different aetiologies of cystic neck swellings, a cystic metastatic tongue squamous cell carcinoma where the primary was detected on random biopsies during panendoscopy and a primary branchiogenic carcinoma. It is important to exclude occult primary before diagnosing primary branchiogenic carcinoma.

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