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Atypical adenolymphoma and glomus caroticum tumour: a rare coincidence

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Abstract. *Atypical adenolymphoma and glomus caroticum tumour: a rare coincidence*. We describe the rare simultaneous appearance of an atypical adenolymphoma with a glomus caroticum tumour on the same side of the neck in a middle-aged man. This case report is the first to describe this coexistence. Due to the atypical, cyst-like presentation of the Warthin's tumour, a final diagnosis was made only after surgical resection and histopathological examination. Both the adenolymphoma and glomus caroticum tumour were successfully removed surgically.

Introduction

We report the case of a 57-year-old man with a Warthin's tumour occurring simultaneously with a glomus caroticum tumour.

Case report

A 57-year-old man presented with a progressive, painless swelling of the left parotid gland for 2 years. The patient had no other complaints. The patient had a 40-pack-year history of cigarette smoking but had quit smoking one month prior. Clinical examination showed a firm, mobile, and non-tender mass in the left parotid region. Bulging of the left pharyngeal wall was observed upon inspection of the mouth with fiber-optic laryngotracheoscopy showing interarytenoid pachydermia.

Consequent ultrasound examination revealed two separate masses on the left side of the neck. The first lesion was a large superficial, well-defined cystic structure. The second lesion was a smooth and hypervascular mass localized at the carotid artery bifurcation with characteristics

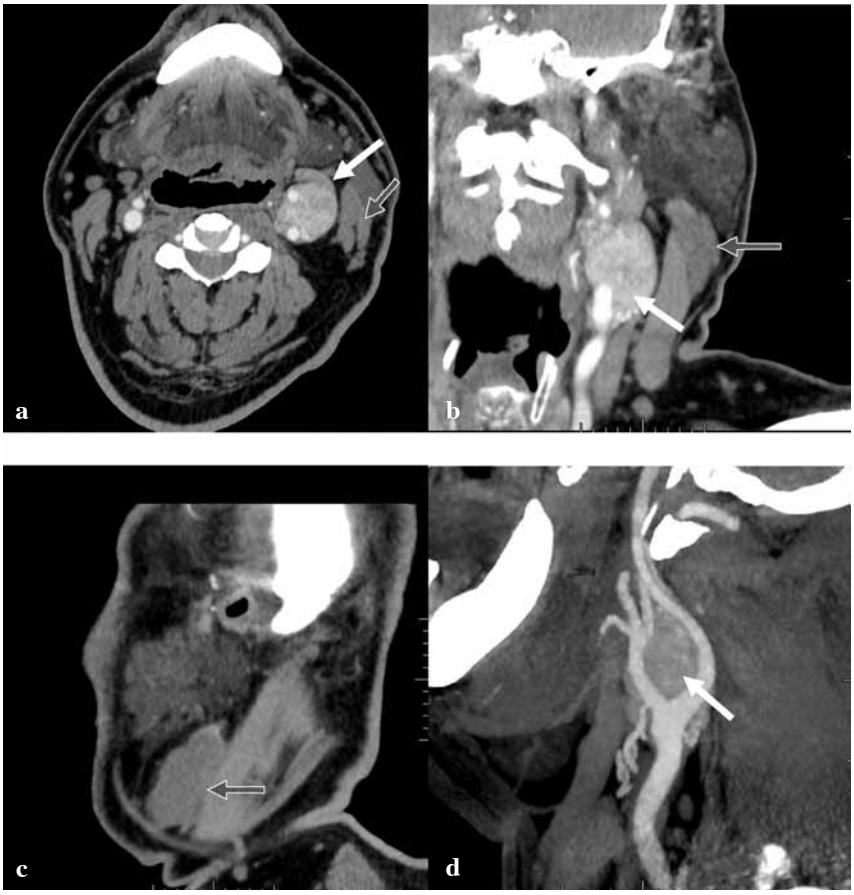

Figure 1
Contrast-enhanced multislice CT of the neck. (a-c) Multiplanar CT reformations show an evident hypervascular mass at the carotid artery bifurcation with imaging characteristics typical of a glomus caroticum tumour (solid white arrow). At the same anatomic level, an oval-shaped, more superficially located and sharply demarcated soft tissue structure is noted adjacent to the lower pole of a normal appearing parotid gland (hollow white arrow). No other anatomic relation with this gland is seen. (d) The maximum intensity projection image further illustrates the splaying of the carotid arteries at the bifurcation, a typical characteristic of glomus caroticum tumours.

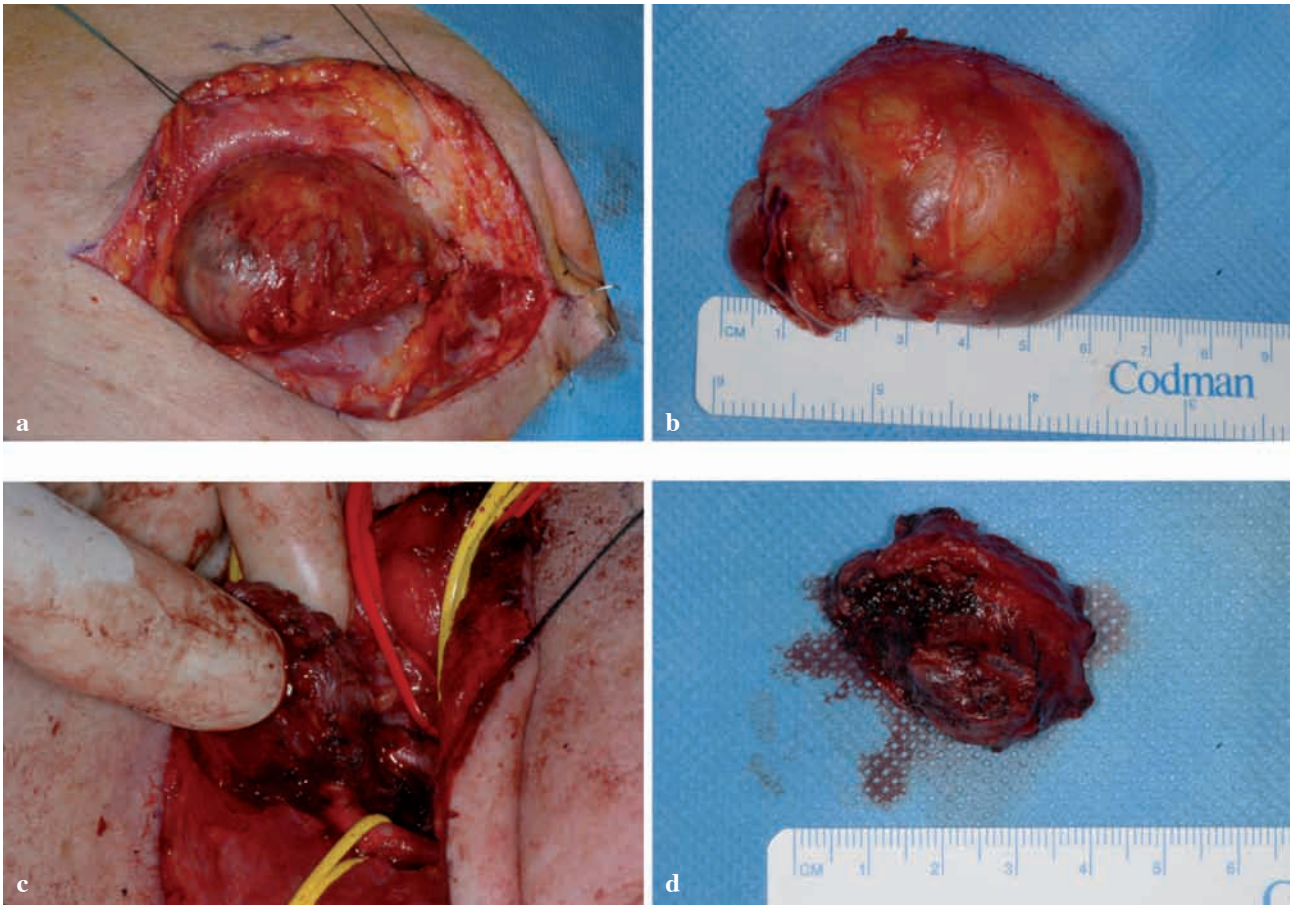

Figure 2
Peri-operative and post-operative macroscopic images of Warthin's tumour (a, b) and glomus caroticum tumour (c, d). a and b show a large, superficial and lobulated cyst, whereas a well-delineated, lobulated, and hypervascular tumour is shown in c and d.

compatible with a glomus caroticum tumour. An ultrasound-guided fine needle aspiration biopsy was performed for the first lesion, revealing few lymphocytes, a majority of which appeared autolytic. This non-specific cytological image was compliant with a benign cyst. No biopsy was performed of the deeper lesion given the hypervascular nature of the presumed glomus caroticum tumour.

Computed tomography (CT) angiography confirmed the two distinct tumours. The superficial structure (37 × 15 × 39 mm) was localized against the outer border of the left sternocleidomastoid muscle and almost completely caudal to the left parotid gland without any obvious anatomical connection to the gland. The structure had a rather low density, suggesting fluid content (Figures 1a-1c). Based on the CT images, cystic lymphangioma was suggested. The lesion at the left carotid artery bifurcation (30 × 29 × 48 mm) was hypervascular and sharply delineated with a location and morphology characteristic of a glomus caroticum tumour (Figures 1a,1b,1d). Additionally, a whole body scan with indium-111 octreotide, a somatostatin analog, was performed, confirming the glomus caroticum tumour as a focal area of high radiotracer uptake in the left neck. No other pathological areas were detected.

The two distinct structures were carefully resected, with complete removal of both lesions (Figures 2a,2c). Histopathological examination of the superficial lesion revealed a lobulated and fluctuating cyst (Figure 2b). The deep lesion appeared lobulated and well-delineated with a red-brownish and very vascular aspect (Figure 2d). Microscopy of the superficial lesion showed a dual structure of enlarged cystic spaces delineated with two layers of eosinophilic epithelial cells surrounded by dens lymphoid stroma (Figure 3). These findings are compliant

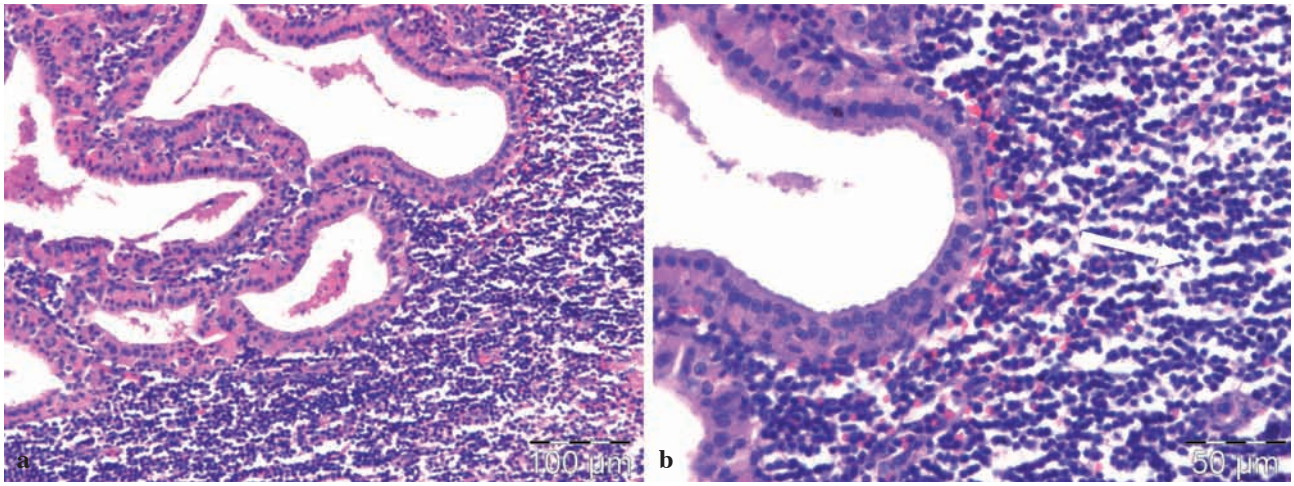

Figure 3
Histological specimen of the cystic degenerated Warthin's tumour showing cystic spaces lined with a double layer of cuboidal to tall columnar eosinophilic epithelial cells and surrounding dens lymphoid stroma (HE).

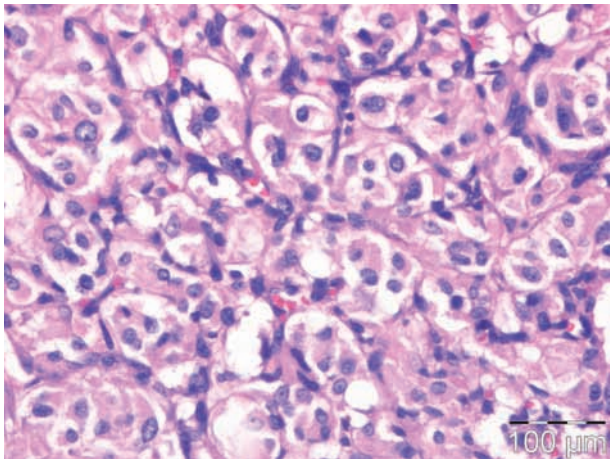

Figure 4
Histological specimen of the glomus caroticum tumour showing a well-developed "zellballen" growth pattern (HE). The "zellballen", or cell nests, consist of tumour cells with supporting or sustentacular cells at the periphery of the cell nests.

with the diagnosis of a cystic, de - generated Warthin's tumour. The deeper lesion appeared as a vascular lesion with a fibrotic pseudo - capsule and a "zellballen" growth pattern, which confirmed the diagnosis of an extra-adrenal paraganglioma or glomus caroticum tumour (Figure 4). The post-operative period was uncomplicated and the patient was discharged from the hospital 3 days later.

Discussion

The simultaneous occurrence of an adenolymphoma (Warthin's tumour) with other neck masses is uncommon. Approximately 12% of patients with Warthin's tumour develop multiple similar tumours, which may be bilateral.[1] However, simultaneous unilateral or bi - lateral involvement of the parotid glands and neck by multiple adenolymphoma has been described,[1,2] though simultaneous occurrence with other tumours of different origin is extremely rare. The co-existence of Warthin's tumour with oncocytoma,[3] MALT-type lymphoma,[4] and pleiomorphic adenoma[2,5] has been reported in the ipsilateral parotid gland. Only three cases of ipsilateral synchronous Warthin's tumour and mucoepidermoid carcinoma have been reported in the literature.[6] Other malignant tumours that have been described to occur with a Warthin's tumour are acinic cell carcinoma, squamous cell carcinoma, adenocarcinoma, adenoid cystic carcinoma, and salivary duct carcinoma.[6] As far as we could verify, the coexistence of an adenolymphoma and glomus tumour has not been described previously.

The two tumours have different etiology and risk factors, as well as a different genetic and embryologic origin (Table 1).[5,7-9] Our patient's smoking habit constitutes a risk factor for both conditions, but no other obvious links were found or considered. None of the

Table 1
Characteristics of Warthin's tumour and glomus caroticum tumour

Characteristic	Warthin's tumour	Glomus caroticum tumour
Gender	male	female (familial form)
Age	50-70 years	45-50 years
Risk factors	smoking	higher altitude, smoking, COPD
Malignancy	0.3%	5-10%
Etiology	proliferation of salivary gland epithelia enclosed in a lymph node (hypothesis of heterotopia)	chronic hypoxic stimulation (sporadic form), genetic predisposition (familial form)
Treatment	surgery	surgery

Table 2
Use of different diagnostic modalities in the pre-operative diagnosis of adenolymphoma (Warthin's tumour)

Diagnostic modality	Characteristics of adenolymphoma
Ultrasound	– evaluation of location, structure, and vascularity of the lesion – well-defined, anechoic mass – often at lower pole of parotid gland – multiple septations possible – thick intratumoural fluid may give an inhomogeneous echogenic pattern
CT/MRI imaging	– evaluation of multicentric, bilateral, or extra-parotid location – well-circumscribed mass – usually 3-4 cm in diameter – complex internal architecture with solid and cystic components
Sialography	repression of the normal structure of the salivary gland
Scintigraphy with radio-labeled 99m Tc-pertechnetate	higher uptake ("hot spot") and retention by lesion compared to surrounding salivary gland tissue
Fine-needle aspiration cytology (FNAC)	flat sheets of oncocytes and lymphocytes scattered in a cystic background

pre-operative diagnostic techniques showed obvious characteristics of an adenolymphoma. Even in a retrospective review of the CT examination, no clear connections with the parotid gland were visualized. Finally, the Warthin's tumour in this case report is remarkable due to its atypical presentation with cystic degeneration. These findings confirm that the pre-operative diagnosis of an adenolymphoma can be challenging.[10,11] Sex, age, and clinical presentation can provide an initial indication.[12] Imaging techniques are helpful in the diagnosis of adenolymphoma, but none can fully exclude the presence of malignancy or provide certainty about the diagnosis (Table 2).[12-15] Final diagnosis is often only made based on histopathological findings after surgical resection, as in our case (Figure 3).[11,12]

Conclusion

Simultaneous occurrence of an adenolymphoma (Warthin's tumour) and glomus caroticum tumour is rare and not previously described in the literature. Whether this association is a coincidence or an unknown common etiology exists remains to be established. Pre-operative diagnosis of a Warthin's tumour with conventional imaging techniques can be challenging, especially with an atypical presentation, as in this case. Warthin's tumour should be included in the differential diagnosis of cystic neck masses, even when imaging does not reveal an obvious communication with the parotid gland. Complete surgical resection is the treatment of choice for both tumours, and the overall prognosis is good.

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