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Ectopic Warthin's tumour presenting as a neck mass

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Abstract. *Ectopic Warthin's tumour presenting as a neck mass. Introduction*: Warthin's tumour usually involves the parotid gland. However, it can also arise from ectopic salivary tissue in the para-parotid and latero-cervical lymph nodes. *Case report*: We present the case of a 60-year-old man with a 3-month history of a smooth, mobile mass on the right side of the neck. Computed tomography (CT) scanning (coronal, axial and sagittal sections) showed a cystic lesion in the right upper neck without connection to the major salivary glands ($8 \times 4 \times 3$ cm). Complete surgical excision with a transverse neck incision was performed. Histological findings of the specimen revealed Warthin's tumour.
Conclusion: This neoplasm should be included in the differential diagnosis of cystic lesions of the neck. Although rare, it has potential for malignant transformation. Coronal and sagittal CT scans are necessary to accurately localize the tumour and to differentiate the diagnosis from earring lesions of the parotid tail.

Introduction

Cystic tumours of the neck are usually inflammatory masses, lymph nodes with centric necrosis, benign and malignant tumours, or metastases.[1] Malformations are less common in adults, including cystic lesions related to abnormal embryogenesis of the thyroglossal duct,[1] the lymphatic primordial,[1] the branchial apparatus,[1] and ectopic thymus tissue.[2] It should be noted that these cystic masses can also result from a tumour developing within the malformation.[3] Ectopic salivary tissue is a rather rare, but clinically important malformation. Warthin's tumour, or papillary cystadenoma lymphadenosum, is the second most common salivary gland tumour.[1] It is a benign, slow-growing tumour found almost exclusively in the parotid gland or in the periparotid lymph nodes.[4] It can also arise in ectopic salivary tissue in the latero-cervical lymph nodes and, therefore, it should be considered in the differential diagnosis of cystic lesions of the neck.[5] This article presents a case of an ectopic Warthin's tumour in the neck with review of the current literature.

Case report

We present the case of a 60-year-old man who was referred to the ENT outpatient clinic with a 3-month history of a gradually growing mass in his right upper neck. A smooth, mobile neck mass anterior to the left sternocleidomastoid muscle in the carotid triangle was found upon physical examination. There was no palpable enlargement of the cervical lymph nodes or evidence of inflammation. Upon general physical examination, the man was found to be moderately built and well-nourished and to have satisfactory vital signs. The fibroscopic examination of the head and neck did not reveal any primary malignancy. The patient had a history of smoking (one-pack-per-day) for more than 30 years. The patient's family history was negative regarding similar conditions.

Based on the history and clinical examination, the initial diagnostic procedure led to a diagnosis of a benign lesion. The differential diagnosis included a brachial cleft cyst, lymphatic-cystic hygroma, laryngocele, atypical tuberculosis, necrotic cystic lymph node, vascular malformations, and rare conditions such as ectopic Warthin's tumour and ectopic thymic cyst.

An ultrasound examination of the neck was performed that showed a cystic lesion in the right upper neck in close proximity firstly with the parotid, and secondly with the submandibular glands; however, it was impossible to exclude a connection between the cyst and the parotid gland. A computed tomography scan (CT) completed the imaging investigations. The CT scan of the neck included axial, coronal, and sagittal sections (Figure 1) to accurately localize the mass and determine its relationship with the major salivary glands, providing

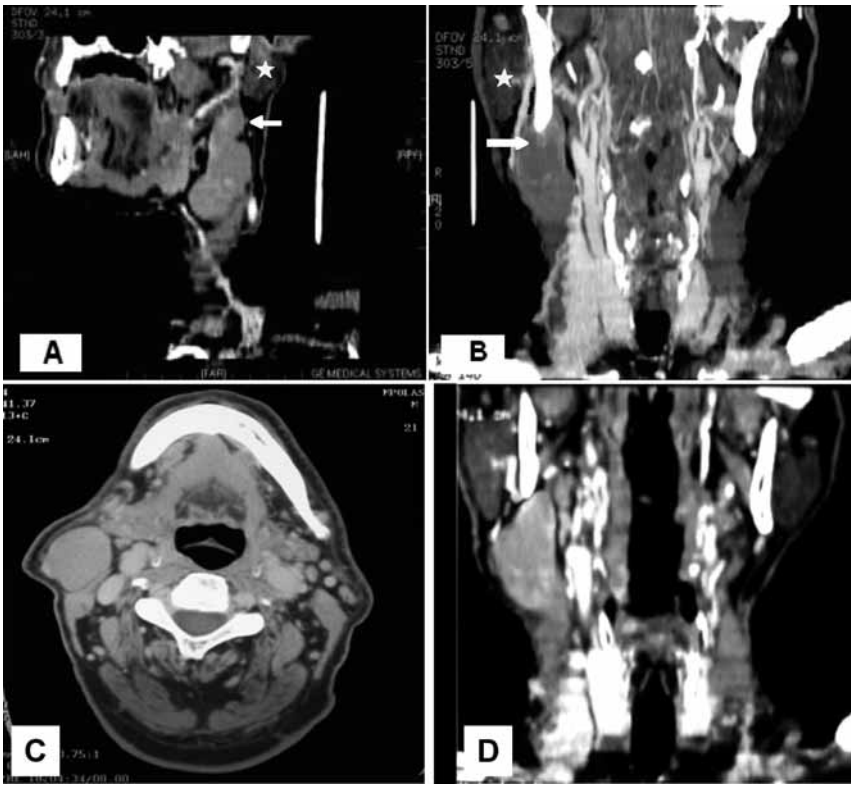

Figure 1
CT scans of the neck (A: sagittal section; B and D: coronal sections; C: axial section) where the extraparotid location of the mass was confirmed. The upper pole of the mass is indicated by the white arrow and the asterisk indicates the parotid tail.

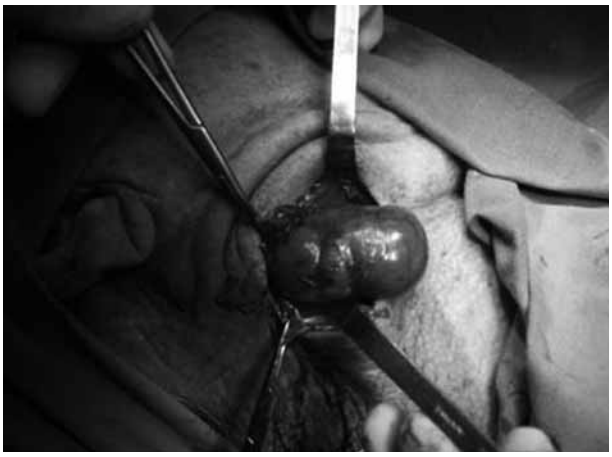

Figure 2
Intraoperative view of the removal of the oval shaped mass ($8 \times 4 \times 3$ cm).

details regarding its extra-parotid origin. No pathological lymph nodes were identified.

The mass was completely excised under general anaesthesia through a transverse neck incision (Figure 2). In the operative field, the mass was found to be encapsulated and was clearly in an extra-parotid location, lying about 1 cm below the parotid tail and 2 cm posterior to the submandibular gland. The specimen was reddish in appearance and measured $8 \times 4 \times 3$ cm.

Microscopic examination of the specimen revealed a mass composed of cystic or cleft-like spaces filled with serous secretion. These spaces were lined with a double layer of epithelial cells that rested on a dense lymphoid stroma bearing germinal centers (Figure 3). The double–cell layer of lining was distinctive, with a surface palisade of columnar cells with abundant fine-granular, eosinophilic cytoplasm. No mitotic activity was observed. The histological findings from the specimen were positive for Warthin's tumour with no evidence of malignant transformation. The patient had an uneventful recovery and he remains in good condition without recurrence one year post-operatively.

Discussion

Papillary cystadenoma lymphadenosum, widely known as Warthin's tumour, tends to occur in older adults between the ages of 60-70 years.[1] It accounts for 4%-15% of all epithelial salivary gland neoplasms, both benign and malignant.[1,6] Typically, Warthin's tumour is an encapsulated, smooth, round lesion with multiple communicating cysts. In 5%-14% of cases, Warthin's tumour is bilateral or even multifocal.[1,6,7] The tumour is almost exclusively located in the parotid gland near the angle of the mandible.[1,4,6] This peripheral tumour location can raise questions about the tumour's origin in some cases (pediculated mass originating from the parotid gland or true extraparotid lesion). Clinicians should remain aware that the tumour may also develop

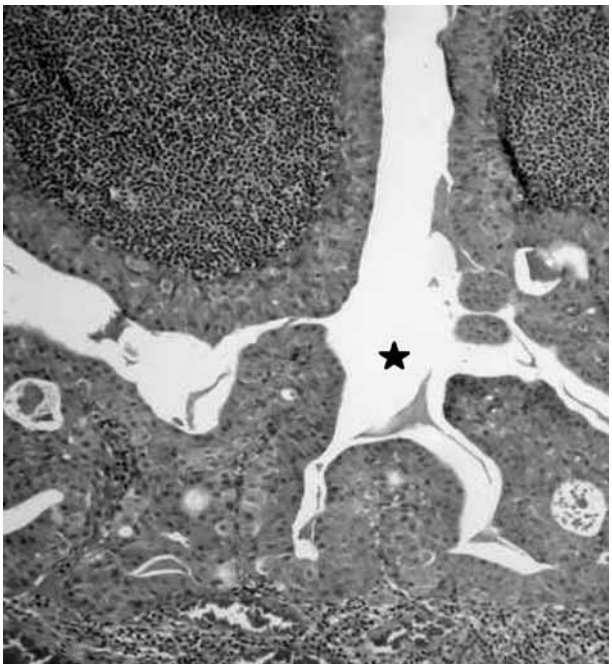

Figure 3
Microscopic examination of the specimen shows cystic spaces (asterisk) surrounded by a double layer of epithelial cells and lymphoid stroma (H&E stain, × 60).

in ectopic salivary tissue, since 8% of the Warthin's tumours arise from an extraparotid location.[5,7] Cervical lymph nodes are the best known and most widely accepted ectopic location.[5,7] Extraparotid Warthin's tumour is believed to be a consequence of the late encapsulation of the parotid gland during embryologic development. Salivary ducts and acini trapped within intraparotid or extraparotid lymph nodes are believed to be the origin of these tumour locations.[5,7] Snyderman *et al*.[7] reported 14 cases of extraparotid Warthin's tumour over a 22 year period, and all occurred in lymph nodes in the neck. This hypothesis is also supported by a retrospective study by Foulsham *et al*.[8] Although the neck is the usual site of ectopic Warthin's tumour, there are reports of unusual distant extraparotid sites such as the nasopharynx,[9] the larynx,[10] the paranasal sinuses,[10] hard palate,[11] and the parapharyngeal space.[12]

Malignant transformation of Warthin's tumour is very rare and constitutes about 0.3% of all Warthin's tumours.[13] The lymphoid component may evolve into malignant lymphoma,[14] whereas the epithelial component may develop into mucoepidermoid carcinoma,[15] adenocarcinoma,[16] or squamous cell carcinoma.[17]

Masses in the parotid tail can be a source of consternation to radiologists and clinicians. Inaccurate localization may lead to a significant iatrogenic complication. The risk of damage to the marginal mandibular branch of the facial nerve during excision of these lesions can be high unless appropriate measures are taken to achieve facial nerve control. The possibility of facial nerve damage increases if there was a previous infection of the mass. The resulting fibrotic formations may produce adhesions to the facial nerve, making surgery more difficult. There is evidence that these episodes of infections start as intra-tumour infarctions which lead to cystic necrosis, inflammation, and fibrosis in approximately 6% of all Warthin's tumours.[18]

Axial CT is the most common diagnostic imaging method for evaluating a head and neck mass; however, a complete diagnostic investigation for a neck mass should always include coronal, sagittal, and axial images. There are two reasons for confusion on axial CT scans. First, a pediculated mass in the parotid tail may not be surrounded by normal parotid tissue and may not be perceived as arising from the gland. In addition, older patients may have atrophic fat-infiltrated glands so that a mass in the parotid tail has less contrast against the adjacent subcutaneous fat.[19] Thus, a mass in the parotid tail might not be perceived as arising from the gland if wide window settings are not used. The location of a parotid tail mass or a true ectopic salivary gland tumour is better appreciated on coronal and sagittal images of the neck as seen in our case, where localization was less confusing.[19]

Fine-needle aspiration (FNA) is a common modality used to investigate salivary gland lesions prior to surgery. However, the diagnostic value of the procedure depends on the institution and the experience of the cytopathologist.[20] In addition, FNA may be complicated by hemorrhage and infection.[20] In a tertiary hospital, patients with Warthin's tumour were found to have a significantly higher risk for FNA induced parotitis because of the nature of the tumour.[21] Specifically, the poor blood supply

of the tumours makes them more susceptible to infarction and inflammation. Clinicians in centers with laboratory personnel who are inexperienced with salivary gland tumours should weigh the risk of inflammation when debating the necessity for routine FNA in a suspected Warthin's tumour.

This type of tumour is highly associated with cigarette smoking.[22,23] This correlation seems to be positive, even for extraparotid sites as seen in our case. The risk of developing Warthin's tumour is two times higher in smokers than in the general population.[22,23] Warthin's tumour traditionally arises in men, but the incidence in women is steadily rising.[22] This change may be due to the growing population of female smokers. It is also reported that smokers who have Warthin's tumour smoked more heavily than did smokers with other salivary gland tumours.[22]

Optimal treatment of Warthin tumour remains somewhat controversial. According to Batsakis,[13] who has written extensively about Warthin's tumour, they are "generally regarded as being among the most innocuous of salivary gland tumours." However there is a possibility of malignant transformation with Warthin's tumours. Considering that a definitive diagnosis requires a tissue sample and that imaging techniques and fine-needle aspiration lack the necessary accuracy for differentiating benign from malignant lesions, a complete surgical excision-biopsy seems to be the most appropriate choice of treatment.

Conclusion

Warthin's tumour should always be included in the differential diagnosis of cystic lesions of the neck. Although rare, it has the potential for malignant transformation. Coronal and sagittal sections of CT scans are helpful to accurate localize the tumour and the confirmation of its origin from the parotid tail or the extraparotid lymph nodes.

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