Case report

Extraparotid Warthin's tumour: usual tumour adjacent to usual site

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ABSTRACT
Lateral neck swelling is a common presentation in ORL practice. The complexity of the neck structures warrants a thorough examination and investigation to narrow down the differential diagnosis. Neoplasm need to be ruled out especially if the neck swelling present in an adult. We report a case of a lateral neck mass being treated as lymphadenopathy before the patient defaulted follow up. The patient represented after 3 years and investigations performed. FNAC revealed sialadenosis but radiologically and intra-operatively consistently showed the mass located outside parotid gland. The final diagnosis of Whartin's confirmed after the HPE evaluation. The diagnosis dilemma is discussed.

Keywords: Lateral neck swelling, warthin's tumour, extra parotid

Introduction
Neoplasm is the most common diagnosis of a lateral neck mass presenting in an adult. Rapid growth over days usually indicates an inflammatory process whereas slow growing mass is more suggestive of neoplastic origin. A benign salivary glands tumour usually present as unilateral, mobile and slowly growing in size over months to year. Location wise, a parotid gland tumour particularly presented with a painless mass anterior to the auricle. Occasionally the mass can be at the tail of parotid and this will be confirmed by means of radiological and intra-operative findings.

Case Report
A 38 year old Malay gentleman, a chronic smoker, complaint of right neck swelling was progressively enlarged over the past 3 years. The swelling was discovered when he experienced right ear pain. During that time, a small swelling was noted at angle of right mandible, the size and location of the swelling suggestive of lymph node, and the diagnosis of lymphadenopathy were made by the attending medical officer. Unfortunately, the patient defaulted follow up before proper investigation as initiated. Since then, the swelling was gradually increased in size. It was not associated with pain. He denied history of dysphagia, odynophagia, facial asymmetry, and any constitutional symptoms. There was no history of dental extraction or contact with tuberculosis patients.

Clinically, a firm, non-tender lesion, measures 4 x 5 cm at right level 2, below angle of mandible with smooth surface. The lesion was mobile in all directions without signs of inflammation on the overlying skin. The earlobe and tragus were not displaced (Figure 1). Examinations of nasopharynx, oropharynx, larynx and hypopharynx revealed normal findings.

Computed tomography (CT) scan of neck revealed a well-defined rounded enhancing mass measuring 3.0 x 3.0 x 4.3 located just inferior to the right parotid gland with poor plane of demarcation with the gland, suggestive of the origin of the swelling (Figure 2). Clear fat plane with the adjacent structure namely sternocleidomastoid, submandibular gland and masseter muscle. Both parotid and submandibular glands are normal. Parapharyngeal and infratemporal fossae are unremarkable.

Cytology examination reported as sialadenosis due to presence of cluster of normal acinar cells without features of pleomorphic adenoma and carcinoma. Subsequently, a superficial parotidectomy was performed under general anesthesia. Intraoperatively, the mass was noted to have a well-defined lesion at the region of tail of parotid, with no extension or connection with the parotid gland. The lesion measuring 5.5 cm x 3.0 cm, oval in shape, smooth and non-lobulated surface (Figure 3&4) and the lesion was excised completely.

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Histopathological examination exhibited well defined encapsulated tumour with papillary structure lined by few layers of oncocytic cells. The papillae core is composed of lymphoid tissue, forming varying sizes of lymphoid follicles with germinal centers, giving the final diagnosis of right extra parotid Warthin's tumour.

Discussion

Neck swelling is a common presentation in otorhinolaryngology clinic. Owing to the complexity of neck structures, the differential diagnosis is long listed. Besides the clue from the anatomical location, the lateral neck swelling can be classified into benign or malignant and congenital or acquired lesions based on history and clinical examination findings. The investigations which include tissue aspiration for cytology and radiological evidence will provide supportive evidence however the ultimate diagnosis is relied upon histopathological examination of the excised mass.

Primary salivary neoplasm appears benign which likely to be pleomorphic adenoma, the most frequently found salivary gland tumour of the parotid gland. Warthin's tumour should be considered as it is the second most common. Warthin's tumour almost always in connection with the parotid gland almost 95% of cases,1 very often it was found in the lower pole, occupying superficial compartment.2 However in our case, the FNAC turned out as sialadenosis.

Cytological investigation is safe, quick and simple diagnostic procedure in investigating neck mass. The FNAC has 74% diagnostic sensitivity and 99% specificity with overall accuracy of 94% for benign salivary gland tumour.3 When cytology shows more abundant acinar tissue sialadenosis may be considered, as demonstrated in our case. However, this diagnosis is usually based on assumption that the
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Acinar tissue is representative of the tar geted lesion. Therefore, sialadenosis is not specific diagnosis and typically represent a missed lesion.4

To further assess the clinically suspected involved gland, imaging investigation is crucial in order to define the exact location, intra-glandular or extraglandular, malignant features, extension and inva

sion and metastases. CT scan has 100% sensitivity and 42% specificity to identify malignant lesion preoperatively.5 Based on CT scan, no malignant features of carcinoma noted such as infiltrative border, necrotic changes, haemorrhage, fibrosis and calcification. This narrow down the differential diagnosis to pleomorphic adenoma and Warthin's tumour as both has similar characteristics in CT scan that are well-defined, homogenous mass. However, 90% of pleomorphic adenomas involve superficial lobe and demonstrate female predominant. While Warthin's tumour which is typically seen in the tail of parotid with male preferences and related to smoking, the most likely possible diagnosis in this preoperatively is Warthin's tumour.

Treatment of Warthin's tumour or any other benign parotid lesion is superficial parotidectomy .6 In our case, as the mass was very well encapsulated with clear plane with parotid tissue, we opted for excision rather than going deep for superficial parotidectomy. For such 'extraparotid' tumour, some authors supported the role of enucleation for the tumour which is hanging off the inferior pole of parotid gland.7 In addition, Heller et al concluded that enucleation is the treatment of choice for Warthin's tumour as the recurrent rate is no different with superficial parotidectomy and in addition facial nerve paralysis and Frey's syndrome are almost always avoided.8

As demonstrated in the index case, intraoperatively we confirmed the presence of a well circumscribed, soft mass with feeding vessel arising from tail of parotid gland. Microscopic evaluation of the specimen showed Warthin's tumour makes our final diagnosis of extraparotid Warthin's tumour, combining all clinical, radiological, intra-operative and histopathological inputs. The tumour occurs rarely in extraparotid site, comprising about 8% of all Warthin's tumour with most often located in the cervical lymph nodes.2 Van der Wal et al described 10 cases (12%) of extraparotid Warthin's tumour out of total 83 numbers of Warthin's tumour cases studied. The locations of extraparotid sites mentioned in this study were 30% in laryngeal region, 30% around facial region, and another 30% in oral cavity and oropharynx, 10% were noted in submandibular region.9 Embryonic development of parotid glands makes the idea of extraparotid Warthin's tumour is not widely accepted. However, study done by Aguirre et al suggest that the pathogenesis of Warthin's tumour is initially an edematous epithelial proliferation followed by lymphocytic infiltration.10 This finding suggest that Warthin's tumour are not restricted to intra or paraparotid region as minor salivary gland tissue may be included in lymphoid tissue anywhere in head and neck region.

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