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Peculiar presentation of extraparotid Warthin's tumour

Osobliwa lokalizacja guza Warthina położonego poza śliniankami przyusznymi

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Abstract

Aim of the study: To emphasize the importance of meticulous history and examination as well as the awareness of various possibilities of diagnosis and management. **Case study:** Warthin's tumour is a benign tumour which mostly arises from the parotid gland and rarely involves minor salivary glands. Extraparotid or ectopic Warthin's tumour most commonly involves the submandibular gland. We report a rare case of nasopharyngeal Warthin's tumour with an unusual presentation. A 59-year-old man presented with left submandibular swelling with right-sided nasopharyngeal fullness. Biopsy of the nasopharyngeal mass along with imaging was suggestive of Warthin's tumour. **Conclusion:** We would like to highlight the challenge in diagnosing this entity and discuss its management.

Keywords: Warthin's tumour, salivary gland tumours, nasopharynx

Streszczenie

Cel badania: Celem badania było podkreślenie znaczenia skrupulatnego wywiadu chorobowego i badania fizykalnego, jak również świadomości istnienia różnych możliwości rozpoznania i postępowania leczniczego. **Opis przypadku:** Guz Warthina to zmiana łagodna wywodząca się głównie z przyusznego gruczołu ślinowego, która rzadziej obejmuje inne gruczoły ślinowe. Guz Warthina położony poza obrębem ślinianek przyusznych lub ektopowy najczęściej dotyczy ślinianki podżuchwowej. W pracy przedstawiono rzadki przypadek guza Warthina o niezwyklej lokalizacji. Mężczyzna w wieku 59 lat zgłosił się z obrzękiem w okolicy podżuchwowej oraz uczuciem pełności w obrębie nosogardła po stronie prawej. Wyniki biopsji masy zlokalizowanej w nosogardle oraz wyniki badań obrazowych sugerowały rozpoznanie guza Warthina. **Wniosek:** Autorzy niniejszej pracy pragną podkreślić wyzwania związane z diagnostyką oraz omówić leczenie tej jednostki chorobowej.

Słowa kluczowe: guz Warthina, guzy gruczołów ślinowych, nosogardło

INTRODUCTION

Warthin's tumour is the second most common benign tumour of the parotid gland following pleomorphic adenoma. It is also known as adenolymphoma or papillary cystadenoma lymphomatosum⁽¹⁾. It mostly occurs in men in 5th to 7th decade of life and can be multiple and bilateral. However, Warthin's tumour has been reported to occur in the minor salivary gland⁽²⁾. Extraparotid or ectopic Warthin's tumour, albeit rare, has been reported to involve the submandibular gland, cervical lymph node, lip, cheek, tongue, hard palate, lacrimal gland and larynx.

CASE REPORT

A 59-year-old man with underlying hypertension and diabetes mellitus type 2 presented with a 3-week history of painless left submandibular swelling. According to the patient, neck swelling was slowly increasing in size despite being treated previously with oral antibiotics prescribed by a private clinic. Otherwise, there were no shortness of breath, dysphagia or odynophagia. No prior toothache or history of trauma or foreign body ingestion. The patient also had no recurrent nasal or ear symptoms. Further

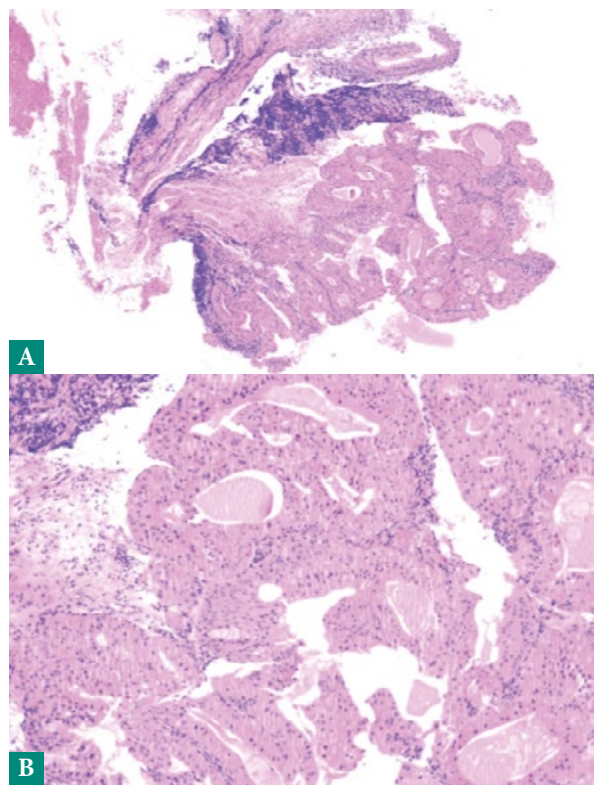


Fig. 1. Nasopharyngeal lesion shows classical histopathological pattern for Warthin's tumour; multiple cystic spaces lined by two epithelial layer, oncocytic luminal cells and tall columnar cells with lymphoid component in the stroma. A. Haematoxylin-eosin, magnification 4 \times . B. Haematoxylin-eosin, magnification 10 \times

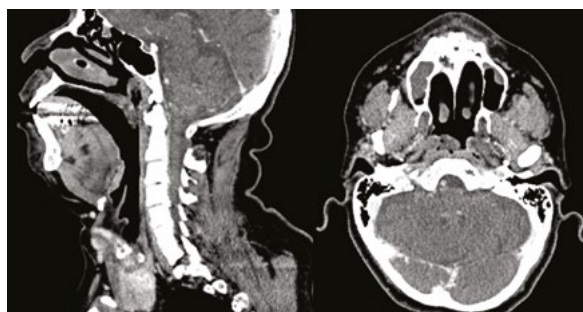


Fig. 2. Computed tomography of the neck revealing non-enhancing nasopharyngeal soft tissue lesion

history revealed a strong family history of malignancy as patient's father had colorectal cancer and his mother suffered from cervical cancer. Other than that, there were no constitutional or B symptoms.

On examination, the patient appeared comfortable, not pale or septic looking. Neck examination revealed left submandibular swelling measuring 2 \times 2 cm, smooth surface, firm, ballotable and not tender. No other neck mass or lymph node were palpable. We proceeded with flexible nasopharyngolaryngoscopy, which revealed obliteration over the right nasopharynx, particularly over the fossa of Rossenmüller. After obtaining consent, biopsy was performed under local anaesthesia along with fine needle aspiration of the left submandibular mass. To our surprise, biopsy from the nasopharynx showed tumour tissue composed of cystic spaces of various sizes lined by two epithelial layers, oncocytic luminal cells and tall columnar cells with lymphoid component of stroma suggestive of Warthin's tumour (Fig. 1). Simultaneously, computed tomography of the neck was ordered which revealed non-enhancing nasopharyngeal soft tissue lesion (Fig. 2). Fine needle aspiration of the neck swelling turned out to be cervical lymphadenopathy. The patient was counselled for surgical intervention of the nasopharyngeal Warthin's tumour; however, he refused and subsequently defaulted our follow up.

DISCUSSION

Warthin's tumour was first described in 1895 by Hildebrand as a type of lateral cervical cyst, which was then described as tumour papillary cystadenoma by Albrecht and Arzt. However, Aldred Warthin reported 2 cases of papillary cystadenoma lymphomatosum manifesting as a slow growing benign neoplasm in 1929⁽³⁾.

Warthin's tumour is a benign tumour of the salivary glands, which arises mainly from the parotid gland and accounts for about 6–10% of all parotid tumours. However, there are a few reported cases of Warthin's tumour arising from other salivary glands. Warthin's tumour has male predilection and is commonly found in individuals aged between 50 and 70 years with a peak incidence the 6th decade of life⁽⁴⁾. Our patient was also a male in his 6th decade of life.

Despite numerous theories on its origin, the most notable one states that remains of ectopic salivary duct epithelium in the lymph nodes of the parotid gland during ontogeny leads to this entity. Encapsulation of the parotid gland occurs later than that of the submandibular gland, and the parotid gland develops with loose capsule. This allows for the invasion of lymphoid tissue in the parotid gland^(5,6). There are a lot of minor salivary glands and lymphoid tissue in the nasopharynx, and the presence of ectopic salivary duct epithelium may explain Warthin's tumour in the nasopharynx as in our case.

Histologically, Warthin's tumour is composed of a double layer of columnar or cuboidal eosinophilic epithelial cells lining a cystic cavity with lymphoid tissue present in the stroma⁽¹⁾. Surgical approaches for excision of tumour may vary. Small sized tumours can be excised endoscopically and large size tumours may need transpalatal approach or pterygoid route, as reported by Yumoto et al. for extensive tumour⁽⁷⁾. However, each case should be evaluated separately, considering the patient factor, environmental factor and surgeon factor. The risk of Warthin's tumour recurrence is low after complete surgical excision (2–5%)⁽¹⁾.

CONCLUSION

Warthin's tumour is a benign tumour which rarely involves minor salivary glands, such as in the nasopharynx. Careful physical and endoscopic examination is recommended for patients presenting with a neck lump. Transnasal endoscopic biopsy should be considered if the lesion is suspicious.

Conflict of interest

The authors do not report any financial or personal connections with other persons or organisations, which might negatively affect the contents of this publication and/or claim authorship rights to this publication.

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