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CASE REPORT

Extraparotid Warthin's Tumor: A Diagnostic Dilemma

Sudharshan, GK Vivek, Aritra Gosh, Donna Bhattacharya

ABSTRACT

Warthin's tumor commonly occurs in parotid gland. Sometimes it has an extraparotid origin like neck, palate, etc. Here we present a case of a neck swelling which created a diagnostic dilemma to be a branchial cyst clinically but later came out to be Warthin's tumor.

Keywords: Warthin's tumor, Extraparotid location, Parotid neoplasm, Surgery.

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INTRODUCTION

Warthin's tumor also known as papillary cystadenoma lymphomatosum is the second most common benign neoplasm of the parotid gland, accounting for 5 to 14% of all parotid neoplasms. Commonly it presents as a slowly growing painless nodular mass of the gland, firm or fluctuant to palpate with a male preponderance (10:1) in their 5th to 6th decade of life.

Pathologically, as the name implies, the tumor grossly appears as an encapsulated mass with cystic and solid areas commonly identified in the lower pole of the parotid gland near the angle of the mandible, 7 to 10% of these tumors can be bilateral while multifocality and recurrence are found in 2% of cases[1-4] Surprisingly, these Warthin's tumors may also be located outside the parotid gland in 8% of cases, which mimic the swellings arising from cervical lymph nodes, the submandibular gland, cystic lesions like branchial cyst or any other lateral neck swelling. These are the cases which may create some confusion in determining the origin of the lesion.[5]

CASE REPORT

A 65 years old male presented with a swelling in the upper part of left side of his neck since 1 year (Fig. 1). The swelling was nontender, not associated with any sudden increase in size, cough, dysphagia or change of voice. There were no associated constitutional symptoms like fever, malaise or loss of weight or appetite. The patient had hypertension and diabetes since last 20 years and is a smoker since last 30 years. There is no previous history of tuberculosis for the patient and his family.

On examination a 5 × 5 cm nontender, soft to firm swelling in the left upper neck, behind the angle of mandible and in front of anterior border of left sternocleidomastoid muscle was found. It was mobile transversely without any overlying skin changes. No other swelling was present in the neck and there was no clinical evidence of cervical lymphadenopathy. The spine and oral cavity examinations were within normal limits. Initial investigations for confirmation of diagnosis included an FNAC, which revealed smear showing cohesive clusters of mature squamous cells along with columnar epithelial cells on background of cystic macrophages, proteinaceous material and scattered inflammatory cells suggestive of an inflamed branchial cyst. Based on the above findings surgical treatment was planned for the patient and in order to determine the origin and plane of the lesion a CT scan of neck was suggested (Figs 2 and 3).

Based on the reports the treatment plan was surgical excision of the lesion and in view of its origin from the left parotid gland, superficial parotidectomy was planned.

The preoperative workup which included routine complete blood investigation, which were within normal limits and chest X-ray and ECG were normal.

At surgery, a superficial parotidectomy approach was made. But it was found that the lesion was completely separate from the lower pole of the left parotid gland and was lying deep to platysma, behind the mandibular ramus and in front of the anterior border of left sternocleidomastoid muscle. The lump was excised with ease through a clear plane of separation from the left parotid gland (Fig. 4).

The histopathology report revealed a 3.8 × 3.0 × 1.5 cm multiloculated cystic tumor with the solid core area showing

Fig. 1: Preoperative picture of patient

vague papillary formation. The cystic spaces and papilla were lined by oncocytic and eosinophillic abundant cytoplasm, oval to round nuclei and filled with mucin and interspersed foamy macrophages, the impression being a Warthin's tumor (Figs 5 and 6).

The postoperative period was uneventful and the patient is coming for regular follow-ups (Fig. 7).

DISCUSSION

The first description of adenolymphoma was done by Hildebrand in 1895 and in 1929 Warthin published a series of so called papillary cystadenoma lymphomatosum leading to the well known term 'Warthin's tumor'. Cystadenolymphomas are the second most frequent lesions of the parotid gland. Due to their benign clinical behavior, low rates of recurrence and malignant transformation they were classified as tumor like lesions. There are several theories to explain the origin and the development of these tumors. A heterotopia of salivary tissue during embryogenesis is the most likely explanation for the origin of these tumors in the upper neck and periparotideal region. During the embryogenesis of the parotid gland, epithelial cells from the oral mucosa happen to penetrate into lymphocyte-rich tissue. The late encapsulation of the parotid gland explains the occurrence of intraparotideal lymph nodes and heterotopic salivary gland remnants entrapped in the parotideal lymph nodes. According to this theory, Warthin's tumors have their origin in these epithelial inclusions.[6]

This hypothesis puts Warthin's tumors in vicinity of lateral cervical cysts, which are thought to originate from proliferating heterotopic inclusions of tonsillar tissue in cervical lymph nodes. In 1967 a relationship between a defective closure of the sinus of the His of the branchial apparatus with heterotopic salivary tissue was suggested by Youngs and Scofield as a possible reason for heterotopic salivary tissue of the lower neck. Other authors propose that aberrant salivary tissue of the pharyngeal pouch may migrate downward with the thyroid and parathyroid gland. Moreover, this hypothesis is further supported by the occurrence of tuberculosis, metastases and malignant

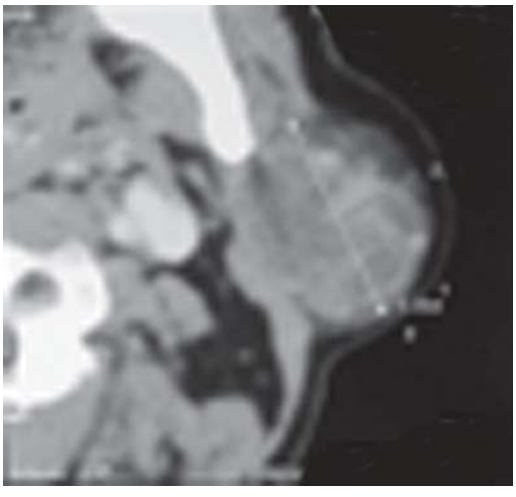

Fig. 2: CT scan showing lump adjacent to parotid gland

Fig. 4: Gross specimen of the excised lesion

Fig. 3: CT neck-coronal section showing lesion in relation to left parotid gland

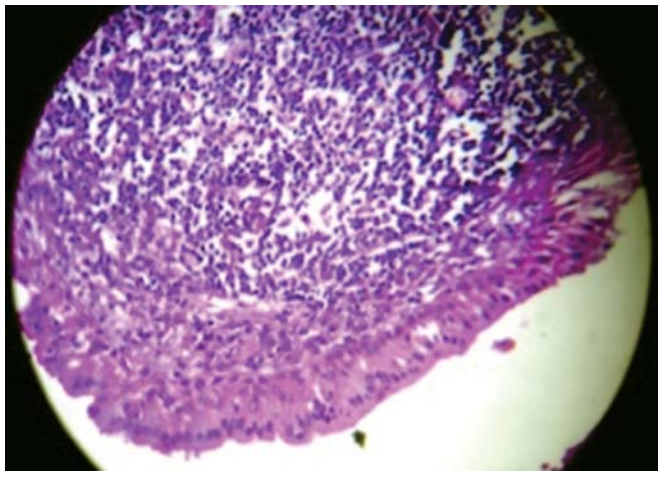

Fig. 5: Photomicrograph showing Warthin's tumor (H&E)

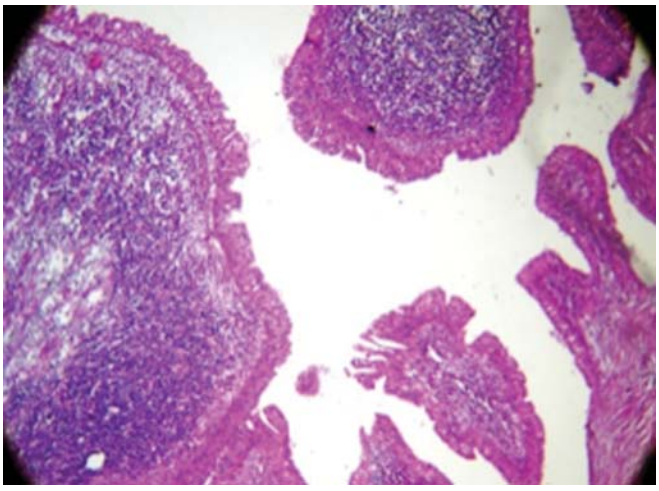

Fig. 6: Core area with vague papillary formation (H&E)

Fig. 7: Postoperative picture of patient

lymphomas in the lymphoid stroma of those tumors. The extraparotid location and multicentric nature of these tumors can be explained by the last mentioned hypothesis.

Warthin's tumor which is exclusively found in parotid gland also has certain other sites of occurrence like lateral neck, palate, upper lip, nasopharynx,submandibular gland, sinuses and lacrimal gland.[7]

Synderman et al have reviewed 14 cases of extraparotid Warthin's tumor.[8] Three patients had lesions discovered in periparotid tissue after superficial parotidectomy for intra-parotid Warthin's tumor. One presented later with a contralateral, clinically evident cervical mass. Four patients presented clinically with a history of an asymptomatic cervical mass. Six patients had ectopic Warthin's tumor discovered incidentally on pathologic examination of neck dissections for other primary head and neck malignancies.

In our case the patient presented with a left sided upper neck mass making it little difficult to assess it as a swelling from the parotid gland. Rather, the clinical differential diagnosis of this lesion would include a variety of benign or malignant, solid or cystic tumors of the cervical area such as branchial cleft cyst, lymphoid hyperplasia, head and neck malignancies with neck metastasis, lipoma, lymphangioma and subcutaneous tumors.

So the only way to get the correct diagnosis is by histopathology which will reveal that, besides the epithelial components these tumors will have a stromal component with lymphocytic infiltrates and lymph follicles.

Optimal treatment of Warthin tumor remains somewhat controversial. According to Batsakis,[9,10] who has written extensively about Warthin tumor, they are 'generally regarded as among the most innocuous of salivary gland tumors'. Malignant transformation is very rare and constitutes only 0.3% of all Warthin tumors. Despite this, Yoo et al recommend superficial parotidectomy but recognized that either local excision or simple observation would be appropriate in selected cases. Their preference is based in part on a recurrence rate of 1.8% after local excision. These may not have represented true recurrences, however, because patients often have multiple Warthin tumors they requires complete excision of the affected portion of the gland with uninvolved margins.

However, the treatment of extraparotid Warthin's tumor is surgical excision.[11,12] Local recurrence after surgical excision is rare.

CONCLUSION

Even though treatment may be local excision, diagnosing and differentiation of extraparotid Warthin's tumor at times can be challenging for the surgeon inspite of the various diagnostic modalities available.

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