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Case Report

Surgery Section

A Rare Coexistence of Papillary Carcinoma, Warthin's Tumour and Oncocytosis

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ABSTRACT

This is the rare case where bilateral parotid glands, bilateral submandibular glands; both lobes of thyroid gland were involved synchronously. Patient was operated for bilateral superficial parotidectomy, bilateral submandibular gland excision and total thyroidectomy. Histopathology report suggested oncocytoma of bilateral parotid glands and one submandibular gland, nontoxic multinodular goiter of thyroid gland and simple cyst of other submandibular gland. This histopathology report was reviewed and reported as oncocytosis of ipsilateral parotid gland, submandibular gland and thyroid lobe, while solid variant of Warthin's tumour of other parotid gland and papillary thyroid carcinoma of contralateral thyroid lobe. The mass of contralateral submandibular gland was reported as simple salivary duct cyst.

On reviewing the literature we found that this type of association was rarely reported and there may be some histopathological relation between oncocytoma, oncocytosis, warthin's tumour and papillary carcinoma thyroid. Although we do not have any conclusive evidence till date but reporting of more such cases may establish some relation between these entities in future.

Keywords: Bilateral parotidectomy, Multiple neck mass, Oncocytoma

CASE REPORT

A 64-year-old male patient presented in department of head and neck surgery with a mass at right parotid region for one and half months. On examination we found 4×4 centimeters sized right parotid mass, 1×1 centimeter sized right level I(b) node, 1×1 centimeter left lower tail parotid mass, and 3×2 centimeters sized left level I(b) node. There was also 4×2 centimeters sized mass noticed in right thyroid lobe along with 2×2 centimeters sized nodule in left thyroid lobe.

His ultrasound neck was done which was suggestive of enlarged thyroid gland with diffuse heterogenous echo pattern with multiple nodules within it which appeared hypo to iso-echoic. Largest hypo-echoic lesion on left side was measuring 4.0×3.0×2.4 centimeters. Largest echogenic lesion on right side was measuring 3.4×2.0 centimeters, a heterogeneous nodule in isthmus measuring 1.4×.7 centimeters and cystic/hypo-echoic speaks at lower pole of right lobe of thyroid measuring 4.0×3.2 centimeters. Major vessels were free from lesion. Multiple nodular deposits were seen in left submandibular gland with echo-pattern similar to the left lobe of thyroid lesion; largest was 2.0×1.5 centimeters. Similar lesion was seen in the right submandibular gland measuring 1.3×1.1 centimeters and in right parotid gland measuring 3.2×1.8 centimeters.

The Impression was-"lesions in Parotid, submandibular and thyroid gland suggestive of possibility of lymphoma/systemic disease". USG guided trucut biopsy from Left submandibular gland lesion showed possibility of oncocytoma or Warthin's tumour and trucut biopsy from thyroid lesion showed adenomatous nodule or follicular adenoma.

Contrast Enhanced Computed Tomography (CECT) scan showed the diffuse thyroid enlargement more on right side with heterogeneous enhancement, ill-defined nodules within. Right lobe was measuring 15.7×5.4×3.8 centimeters and Left lobe was measuring 13×3.4×3.8 centimeters. Isthmus was measuring 1.8 centimeters. The largest nodule in right lobe measures 9.5×4.0×3.8 centimeters [Table/Fig-1]. Bilateral parotid glands were bulky, showed few homogeneously enhancing well defined nodular lesions with largest lesion in right parotid gland measuring 5.5×2.7×3.2 centimeters and in left parotid gland measuring 3.1×1.7×1.0 centimeters [Table/Fig-2]. Bilateral submandibular gland appeared heterogeneous with poorly enhancing areas within it. Bilateral inhomogeneously enhancing enlarged submandibular nodes were found largest on right side measuring 1.3×1.1 centimeters and on left side measuring 1.4×1.6 centimeters [Table/Fig-3]. Impression was heterogeneous enhancing thyroid gland with largest nodule in right lobe (Possibility of lymphoma or follicular neoplasm), heterogeneously enhancing bilateral submandibular glands, bulky parotids with few homogeneously enhancing nodular lesions (Possibility of bilateral warthin's tumour or lymphoma).

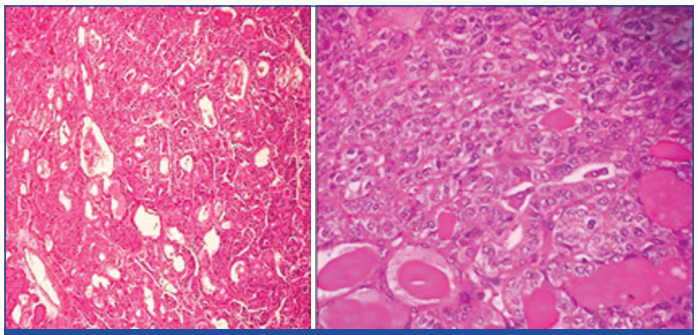
[Table/Fig-1]: Contrast enhanced computed tomography shows multiple Nodules in both thyroid lobes.

Patient underwent Bilateral Adequate Parotidectomy, Total Thyroidectomy, Bilateral level I(b) (submandibular gland) excision. His final histopathology report suggested Oncocytoma of right and left Parotid glands and left submandibular gland with simple salivary duct cyst of right submandibular gland. Thyroid gland showed non toxic multinodular goiter. This biopsy was reviewed and reported as papillary thyroid carcinoma of right lobe [Table/Fig-4,5] while multinodular goiter of left lobe. Solid variant of Warthin's tumour was reported for right Parotid gland [Table/Fig-6,7] and oncocytosis of left

[Table/Fig-2]: Contrast enhanced computed tomography shows bulky bilateral parotid glands with enhancing area in right parotid gland.

[Table/Fig-3]: Contrast enhanced computed tomography Sagital section shows enlarged left submandibular gland.

Parotid gland [Table/Fig-8]. Left submandibular mass was reported as extensive oncocytosis [Table/Fig-9] while benign simple salivary duct cyst was reported for right submandibular salivary gland.

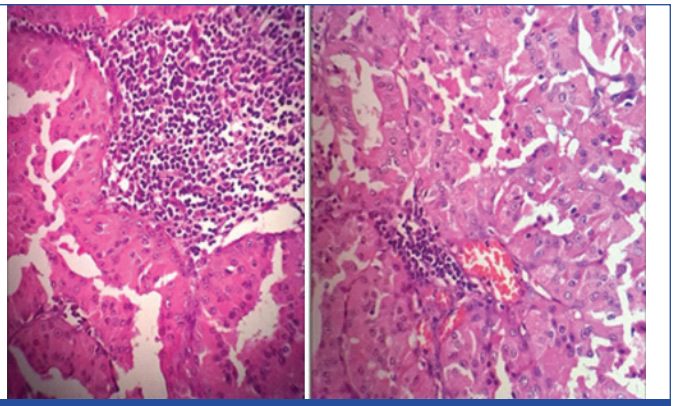


[Table/Fig-4,5]: Right lobe of thyroid-Atypical follicular epithelial cells with nuclear features of Papillary thyroid carcinoma {H&E stains; (10X),(40X)}. [From left to right]

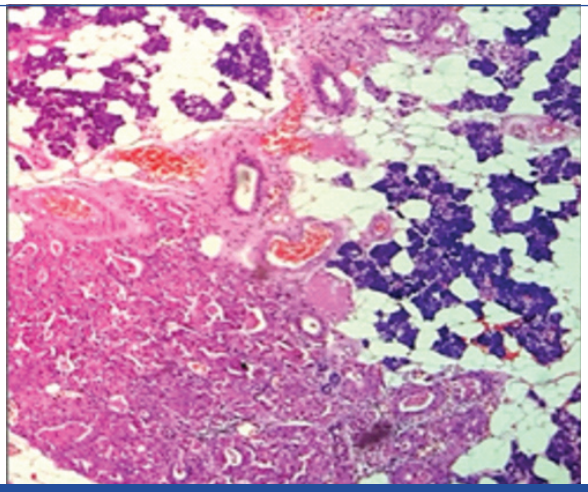
After follow up of one and half year patient developed a nodule at left parotid [Table/Fig-10]. He was operated for the same again. His final histopathology was reported as oncocytoma. Following second surgery patient was regular in follow up till date and he is disease free [Table/Fig-11,12].

DISCUSSION

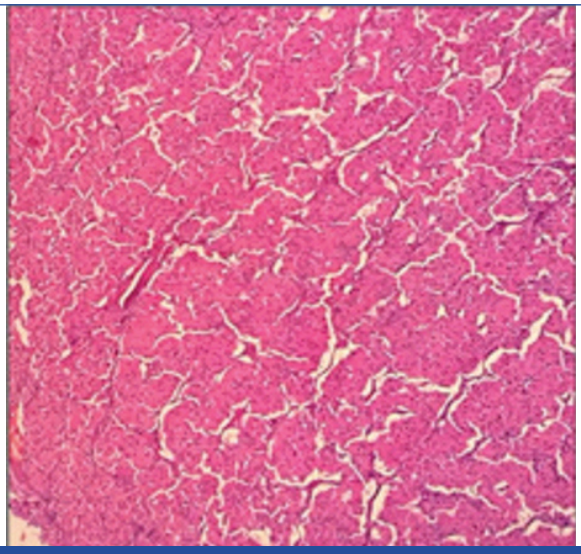
Warthin's tumour was first described by Alderd Warthin's in 1929. Warthin's tumour is benign salivary neoplasm most commonly occur in parotid gland. Histologically, it shows oncocytic epithelial cells in rows with cystic spaces and lymphoid stroma forming



[Table/Fig-6,7]: Right parotid gland nodule-Double layer of oncocytic epithelial cells resting on dense lymphoid stroma consistent with solid variant of warthin's tumour {H&E stains; (40X,40X)}.



[Table/Fig-8]: Left Parotid gland-Sheets of oncocytes (arrow) adjacent to benign salivary acini, consistent with oncocytosis {H&E stain; (10X)}.

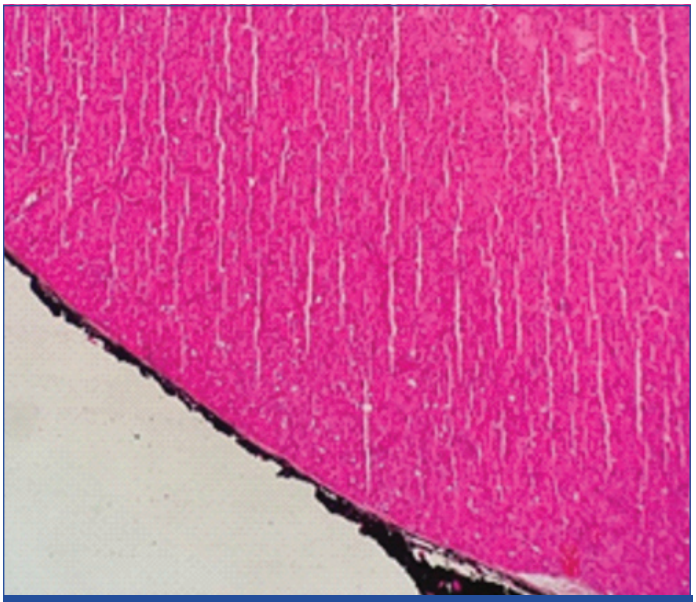


[Table/Fig-9]: Left submandibular gland-solid sheets of benign oncocytes consistent with oncocytosis {H&E stain; (4X)}.

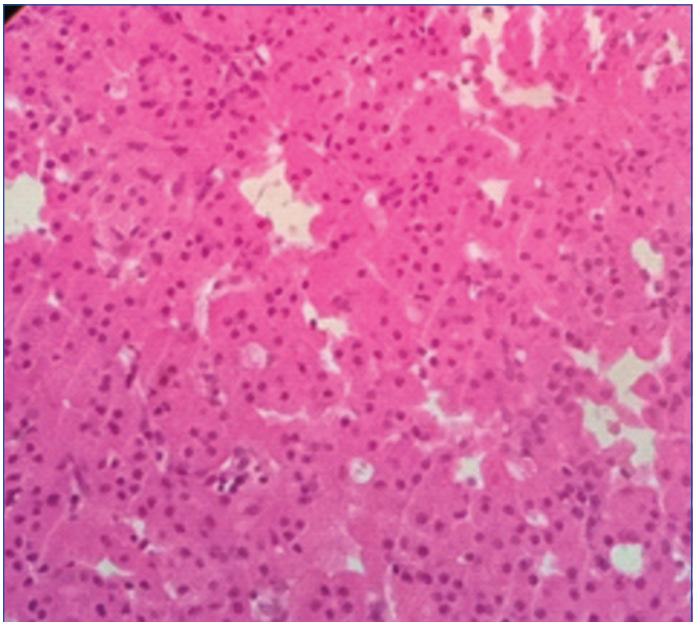
germinal centres [1]. Five to 15 percent cases occur bilaterally, may be multifocal in 6 to 20 percent cases. Rate of recurrence is 0 to 13 percent [1].

Term oncocytoma was given by Jaffe in 1932, they are rare tumours constitute 0.1-1.5% of salivary gland tumours and parotid gland is most common site [2]. Oncocytomas are benign epithelial tumours characterized by oncocytes with eosinophilic granular cytoplasm rich in mitochondria. Oncocytic cells are thought to originate from the transformation of epithelial cells of salivary gland ducts or acini. They occur most commonly in their sixth to eighth decades and are slightly predominant in women. The term oncocyte was given by *HAMPERL* 1931 [2].

[Table/Fig-10]: Contrast enhanced computed tomography shows mass at left parotid gland (recurrence).



[Table/Fig-11]: Final histopathology report of recurrent lesion at left parotid which was reported as oncocytoma-Sheets of oncocytic cells consistent with oncocytoma {H&E stains; (4X)}



[Table/Fig-12]: Final histopathology report of recurrent lesion at left parotid which was reported as oncocytoma-Sheets of oncocytic cells consistent with oncocytoma {H&E stains; (10X)}.

Warthin tumour has histological similarities with oncocytoma like cystic changes and lymphocytic infiltrates which are component of warthin's tumour may be seen in oncocytoma. Moreover, focal oncocytic hyperplasia can also be seen in warthin's tumour. In oncocytoma, Fine Needle Aspiration Cytology (FNAC) shows sheets of epithelial cells and clusters of papillaries forming acinus, minimal lymphoid cells can be seen with prominent single cells while Warthin's tumour shows epithelial cells which are predominantly oncocytic mostly in sheets, rarely papillary fragments, scarce single cells. One can see lymphoid cells mixed with fluid of lymphocytes and debris [3].

The most common malignant thyroid neoplasm is Papillary Thyroid Carcinoma (PTC) which constitutes more than 70 percent of all thyroid neoplasia. Conventional PTC shows papillary architecture with branching papillae are covered by cells with eosinophilic cytoplasm with enlarged nuclei. Many variants of PTC are described in literature [4]. Warthin's like PTC is a rare variant of PTC described by Apel in 1995 in which neoplastic cells with oncocytic changes and clear nuclei, lining the papillary fronds that have lymphatic stroma like stalks. The name given to it is due to its resemblance with Warthin's tumour [5]. Nikiforov YE et al., has observed that solid variant has got poor prognosis and has more incidence of metastasis [6].

There are reported cases of synchronous oncocytoma and warthin's tumour in ipsilateral parotid gland [7] but simultaneous occurrence of warthin's tumour, Papillary carcinoma thyroid and oncocytosis of other glands is rare. There are reported cases [8] of warthin's like variant of papillary carcinoma thyroid which is rare and separate entity but again therir simultaneous occurrence was not reported. This case was unique because of simultaneous occurrence of oncosytosis of multiple glands which indicates some common phenomenon in occurrence in these tumours. Although there is no clear evidence and present data does not support this. As far as treatment was concerned they were treated as individual tumours. Following surgery patient did well and was on regular follow up. After one and half years, he again developed nodule at tail of parotid gland which was operated. Final histopathology was reported as oncocytoma. The recurrence rate of oncocytomas is reported as 20 to 30 percent (after incomplete excision) but malignant transformation is rare [2].

CONCLUSION

This is a rare case of Synchronous occurrence of papillary carcinoma thyroid, warthin's tumour, and oncocytoma. There is diagnostic confusion in oncocytoma and warthin's tumour and we have one variant of papillary carcinoma thyroid i.e., warthin's like PTC. Although all the three entities are pathologically distinct but the above mentioned similarity and simultaneous occurrence led us to review the literature and report this case. Although neither we have literature evidence nor this case concludes similarity between these entities but still these synchronous occurrence indicate something to look for in future.

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