**CASE REPORT**

**Papillary cystic type of acinic cell carcinoma of parotid: fine needle aspiration cytological features of a high grade variant with oncocytic metaplasia**

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**Abstract**

A 60-year-old female developed a right parotid swelling six months after surgery for intra-oral squamous cell carcinoma. Fine needle aspiration (FNA) cytological smears showed dissociated large and small pleomorphic tumour cells with abundant mitoses and oncocytic features. A cytological diagnosis of parotid acinic cell carcinoma (ACC) was made. Histological study of the subtotal parotidectomy specimen showed a papillary cystic variety of acinic cell carcinoma (ACC-PCV). FNA cytological features in this case of ACC-PCV differs from the two previously reported cases in that it showed prominent oncocytic and high grade features and absence of papillary pattern in the cytological smears. ACC-PCV is an uncommon tumour and knowledge of its varied FNA cytological features is important for the diagnosis of this neoplasm.

**Key words:** Parotid, salivary gland, acinic cell carcinoma, papillary cystic, oncocyes, fine needle aspiration cytology

**INTRODUCTION**

Acinic cell carcinomas (ACC) are usually low grade, well differentiated neoplasms that constitute 2.5 to 4% of parotid gland tumours.’ Papillary cystic variant of acinic cell carcinoma (ACC-PCV) is an uncommon tumour that has been mostly reported to occur in younger patients (16-40 years)2,3 as compared to the classic type that characteristically presents in the fifth decade of life.’ Fine needle aspiration (FNA) cytological features of ACC-PCV2,3 are less well recognized as compared to cytological features in classic acinic cell carcinoma.6,8 This report describes FNA cytological features in a case of ACC-PCV of parotid gland occurring in an elderly female who had recently completed treatment for an ipsilateral intra-oral squamous cell carcinoma (SCC).

**CASE REPORT**

A 60-year-old female who was on monthly post-treatment follow-up for a right intra-oral SCC presented with a one month history of a painless swelling in the right parotid region that was rapidly increasing in size. She had presented with intra-oral SCC six months ago and was staged as T2N1. She had no history of tobacco chewing or of radiation exposure to the head and neck in the past. A wide excision of the tumour and a right radical neck node dissection were done. Histopathological study showed features of SCC with excision margins and neck nodes free of tumour. The patient was therefore not scheduled for post-operative radiotherapy.

The swelling over the parotid region was firm, non-tender, mobile and measured 4 cm X 3 cm with well-defined margins. There was no contralateral parotid or any other neck swellings present and examination of the oral cavity ruled out recurrence of SCC. A provisional clinical diagnosis of metastatic involvement of the pre-auricular lymph nodes was made and FNA cytology was requested. The cytological picture suggested a primary parotid adenocarcinoma of possibly acinic cell type and a parotidectomy was advised.

Intra-operatively the tumour was close to the deep lobe of the parotid gland and the facial nerve. The plane between the parotid gland and
the external carotid artery was obscured due to fibrosis occurring after the radical neck dissection. Subtotal parotidectomy was performed with approximately 2 cm margin of normal parotid tissue around the tumour. Histology of the tumour revealed a high grade ACC-PCV with foci of oncocytic metaplasia.

Cytology
Cytological smears stained with May Grünwald Giemsa showed high cellularity with numerous dissociated round and polygonal cells that varied in size from small to intermediate and large sized cells (Figs. 1 & 2). Most of the cells were large with abundant deeply basophilic cytoplasm and eccentric nuclei resembling malignant oncocyttes. Some of the cells showed pink-staining cytoplasmic inclusions of various sizes while others showed cytoplasmic vacuoles (Fig. 2). Nuclei were round and vesicular and were eccentrically placed. In Papanicolaou stained smears, nuclear chromatin was coarse and nuclear membranes showed numerous irregularities and notching (Fig. 1). Some of the cells showed macronucleoli (Fig. 1) and binucleation (Figs. 2 & 3) and a few multinucleated forms were present. Mitotic activity was prominent (Fig. 3) and moderate numbers of lymphocytes were scattered (Figs. 2 & 3). Stains for mucin were negative. No papillary pattern was observed in the cytological smears.

Pathology
The excised parotid gland measured 6 cm x 3 cm x 1 cm and cut section showed a whitish partly necrotic tumour measuring 2.3 cm x 2 cm x 1 cm. Histological sections showed a partly circumscribed tumour composed of round to polygonal cells that were arranged in papillary, alveolar and pseudoalveolar patterns with cystic areas (Figs. 4 & 5). Cytoplasm was abundant and deeply eosinophilic giving an oncocytic appearance to the cells and nuclei were eccentric, round or oval, pleomorphic and vesicular (Fig. 6). Multiple macronucleoli, numerous mitotic figures and binucleated and multinucleated cells were seen as were also cells with bizarre or lobulated nuclei (Fig. 6). Some of the cells showed a rhabdoid appearance (Fig. 6) with pink cytoplasmic inclusions that did not stain for mucin. There was an abundant lymphoid infiltrate around and in between tumour cell groups (Fig. 6). At the periphery of the tumour, normal serous salivary acini and ducts were seen with foci of oncocytic metaplasia. Occasional salivary ducts were cystically dilated and filled with hyaline concretions. The tumour cell's showed immunocytochemical activity for cytokeratin but not for $\beta$100 protein or actin. The margins of the resection were not involved by the tumour. A histological diagnosis of ACC-PCV was made.

DISCUSSION
The variety of appearances of ACC of salivary gland (classic, microcystic, follicular, papillary and papillary cystic)\(^9\) make this neoplasm a diagnostic challenge in FNA cytology. FNA cytological features of classic ACC are well known.\(^5\) Smears show loosely clustered and acinar groups of cells that are generally bland-looking, resembling non-neoplastic salivary acinar cells.\(^2\) Cell dissociation and abundance of bare tumour cell nuclei are some of the features that distinguish well differentiated ACC from normal salivary gland acini.\(^6\) In ACC-PCV, cell dissociation is sparse and acinar structures and bare tumour cell nuclei are missing.\(^3\) On the other hand, numerous small and large papillary groups and monolayered sheets of moderately pleomorphic tumour cells have been described.\(^2,3\)

Prominent oncocytic metaplasia in the present case led to a differential diagnostic consideration of a malignant oncocytoma.\(^1,13\) Oncocytomas of major salivary glands are rare tumours that occur in elderly individuals some of whom have had exposure to radiation.\(^13\) Benign oncocytomas need to be distinguished from non-neoplastic oncocytic metaplasia that is common in the elderly,\(^14\) while malignant variants may be difficult to distinguish from oncocytic metaplasia occurring in ACC, muco-epidermoid carcinoma and malignant mixed tumours.\(^13\) While benign oncocytoma may occasionally have a cystic component with papillae (Warthin-like areas), malignant tumours are predominantly solid.\(^13\) One\(^2\) of two cases\(^2,3\) documenting FNA cytology of ACC-PCV showed oncocytic features in some of the cells; however illustrations from both these reports show relatively low grade nuclear features in the tumour cells in cytological and histological preparations.

The present case of ACC-PCV showed several features that distinguish it from previously reported cases of ACC-PCV. These include occurrence as a second neoplasm in an elderly female with recently treated oral SCC, absence of papillary differentiation in cytological smears coupled with the presence of high grade nuclear features including numerous mitoses.
FIG. 1. Dissociated round and polygonal tumour cells of varying sizes with abundant cytoplasm and eccentric nuclei. Nuclei show coarse chromatin, prominent nucleoli and nuclear membrane irregularities. Pap × 400

FIG. 2. Dissociated large tumour cells with vacuolated cytoplasm and one cell (arrow) showing pink cytoplasmic inclusion. Note lymphocytes in the background. MGG × 800
FIG. 3. Intermediate-sized and large tumour cells with some binucleate forms and inset showing tumour cells in mitosis. Note lymphocytes in the background. MGG x 400.

FIG. 4. Histological section from tumour showing papillary pattern with tumour cells lying dissociated in the cystic area (lower half). H&E x 200.
FIG. 5. Pseudoalveolar pattern of tumour. H&E x 200.

FIG. 6. Oncytoid cells with marked variation in size, pleomorphic nuclei, macronucleoli, binucleated and multinucleated forms and lymphocytic infiltrate. H&E x 400
REFERENCES


