

Case Report

## Recurrent cystadenocarcinoma of parotid: not always an indolent tumor

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### Abstract

Cystadenocarcinoma is a rare malignant tumor, constitutes 2% of malignant tumors of salivary gland known to have an indolent biological behaviour. A 60 years male presented with the history of swelling below right ear. He had similar complaints in the past for which patient had undergone superficial parotidectomy five months back.

Histopathological examination of the excised specimen showed features of Cystadenocarcinoma.

Here we are presenting a case of cystadenocarcinoma which has recurred in a short time even after complete surgical excision with clear margins

**Key words:** Cystadenocarcinoma, salivary glands, papillary growth, recurrent.

### Introduction

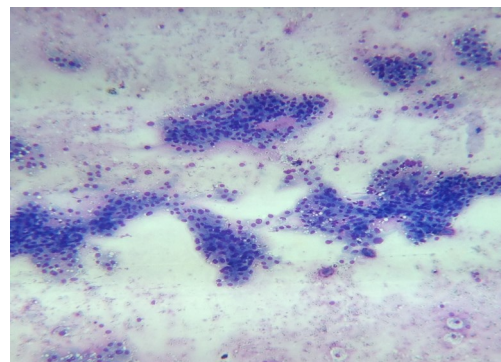
Cystadenocarcinoma is a rare malignant tumor characterized by predominantly cystic growth that often exhibits intraluminal papillary growth. It is a malignant counterpart of cystadenoma, constitutes 2% of malignant tumors of salivary gland known to have an indolent biological behavior.<sup>1,2,3</sup> About 65% of cystadenocarcinoma occur in major salivary glands and the others sites are buccal mucosa, lips and palate.<sup>4</sup>

### Case report

A 60 years male presented with the history of swelling below right ear. He had similar complaints in the past for which patient had undergone superficial parotidectomy five months back. On examination swelling measured 7x6 cm, non-tender, consistency was firm to cystic. Surgical scar was present. Patient underwent excision of lesion with modified radical neck dissection (MRND). On serial sectioning of the specimen gray white nodule measuring

2cmx2cmx1.5cm was seen. Microscopically it was a cystic lesion composed of cells arranged in papillary pattern with thin fibrovascular cores. Cells were showing moderate pleomorphism with vesicular chromatin, prominent nucleoli, and moderate cytoplasm. Some cells are also forming tufts, buds and are arranged in glandular pattern. All margins of the specimen were free from tumor. No lymph nodes retrieved from MRND. After ten months, patient was readmitted with swelling in the right posterior cervical region. Fine needle aspiration of the swelling was positive for malignant cells. Excision of the swelling was done. Histopathological examination of the excised specimen showed recurrent papillary cystadenocarcinoma.

**Figure:1** Aspiration showing papillary cluster with pleomorphic nuclei, moderate cytoplasm. (MGG, 100X)



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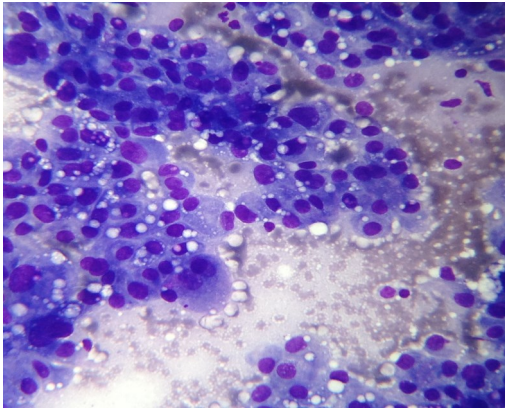
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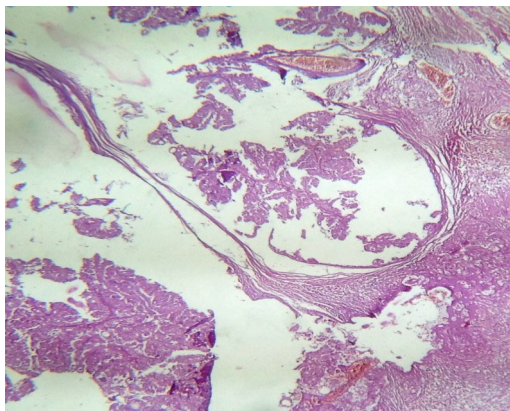
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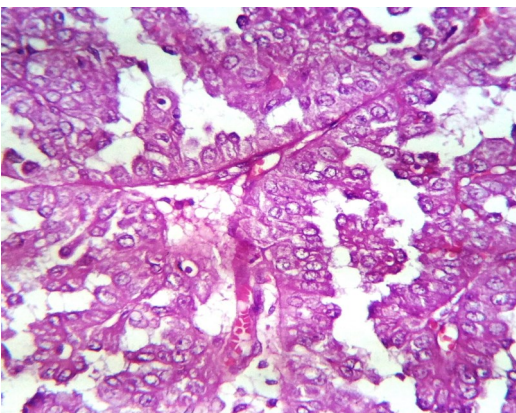
**Figure:2** Papillary structures with pleomorphic nuclei, moderate cytoplasm (MGG, 400X)



**Figure:3** Cystic lesion lined by papillary growth. (MGG, 100X)



**Figure:4** Papillae lined by pleomorphic vesicular nuclei, prominent nucleoli, moderate cytoplasm (H&E, 400X)



## Discussion

Cystadenocarcinoma or papillary cystadenocarcinoma represents the malignant counterpart of cystadenoma, characterized by multicystic growth with intraluminal papillary growth.<sup>5</sup> It is a rare malignant tumor with an incidence of 2% of all the malignant salivary gland tumors. There is no gender preference, and the average age was 57-58.8 years.<sup>2,6</sup> The major salivary gland, parotid is commonly involved in 65%. Other common sites are sublingual gland, buccal mucosa, lips and palate.<sup>2,4</sup>

Cystadenocarcinoma usually presents as a slow growing asymptomatic swelling. The size varies from 0.4 to 6 cm.<sup>2,7</sup> Grossly these tumors are multicystic of variable sizes with focal solid area, sometimes with papillary excrescences. Microscopically, tumors are well circumscribed, with invasive cystic growth pattern and lumen shows varying degree of conspicuous papillary growth. The cells lining the papillae may be medium to large, cuboidal to columnar with scant to moderate cytoplasm, vesicular to coarse chromatin, occasional mitosis may be seen. Tumor must show focal infiltration to the surrounding connective tissue or salivary parancyma.<sup>4,5,7,8</sup>

The most common differentials for cystadenocarcinoma are other malignant cystic lesion of salivary glands. The most common differentials are low grade mucoepidermoid carcinoma and cystic variant of acinic cell carcinoma. Mucoepidermoid carcinoma usually has varied cell types and more solid and acinic cell carcinoma will have focal acinar differentiation.<sup>2,5,8,9</sup>

Currently, complete surgical excision with clear margin is the treatment of choice. Lymphadenectomy is recommended for the patients with lymphadenopathy. Despite of its low grade nature, there are few reports where papillary carcinoma has showed more aggressive behavior.<sup>2,5,8,9</sup> Recurrence of malignancy with lymph node metastasis within a span of one and half year is very rare which occurred in our case.

High grade papillary cystadenocarcinoma are associated with rapid progression and a metastatic potential to cervical lymph nodes. Recurrence and cervical lymph node metastasis has been reported in few cases after 2 to 21 years.<sup>10</sup> Radiotherapy is usually recommended for high grade recurrent tumors or for inoperative patients.<sup>6,11</sup>

## Conclusion

Even though Cystadenocarcinomas are known to have an indolent biological behaviour, these tumors can recur within short time even after complete surgical excision with clear margins.

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