

Case report

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Synchronous sebaceous lymphadenoma with squamous cell carcinoma – case report

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Published: 24 December 2003

Received: 25 August 2003

World Journal of Surgical Oncology 2003, 1:30

Accepted: 24 December 2003

This article is available from: <http://www.wjso.com/content/1/1/30>

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Abstract

Background: Sebaceous lymphadenoma is a rare benign salivary gland tumour of uncertain histogenesis. So is synchronous occurrence of two benign or malignant neoplasms.

Case-report: 68-year-old female presented with right side parotid swelling associated with pain and gradual increase in size. Fine needle aspiration cytology of parotid swelling was suggestive of pleomorphic adenoma. Total conservative parotidectomy was performed and histopathology of the specimen revealed sebaceous lymphadenoma with squamous cell carcinoma.

Conclusions: Sebaceous lymphadenoma and squamous cell carcinoma are two rare benign and malignant neoplasms arising in parotid gland. Synchronous occurrence of these two entities has not been reported.

Background

Synchronous occurrence of two or more pathologically distinct benign and malignant neoplasm in a single organ is rare and accounts for < 0.1% of all salivary gland neoplasms. We report here a rare case of synchronous occurrence of sebaceous lymphadenoma, a rare benign salivary gland neoplasm predominantly seen in elderly females [1] and primary squamous cell carcinoma too is of rare occurrence in parotid gland. The limited number of reported cases of synchronous carcinoma in the salivary glands makes clinical management of these lesions difficult.

Case Report

A 68-year-old woman presented with swelling in the right parotid region of 8 years duration with history of gradual increase in size over last 1 year with pain and tinnitus for

past 2 months. There was no difficulty in opening the mouth and no history suggestive of facial nerve palsy. Medical or family history was not contributory.

Examination showed a non tender swelling with variegated consistency in parotid region raising the ear lobule. Overlying skin showed dilated veins. The swelling was partly fixed to underlying structures and clinically there was no evidence of facial nerve involvement. There were no palpable cervical lymph nodes and haematological and biochemical investigation were within normal limits. A fine needle aspiration cytology (FNAC) was carried out which was suggestive of pleomorphic adenoma. With a diagnosis of pleomorphic adenoma patient underwent parotidectomy.

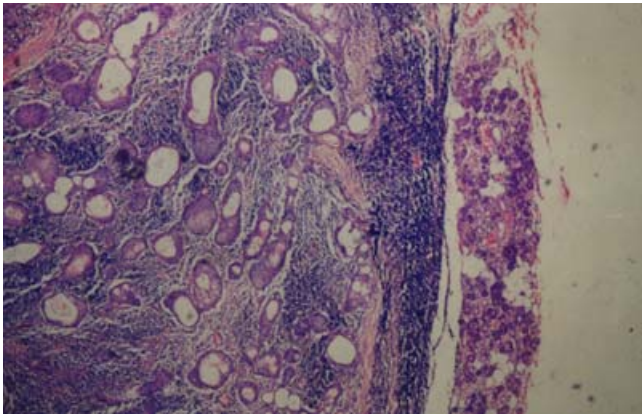


Figure 1
Photomicrograph showing partially circumscribed neoplasm composed of numerous duct like structures, cystic spaces mixed with lymphoid stroma (Haematoxylin & Eosin \times 40)

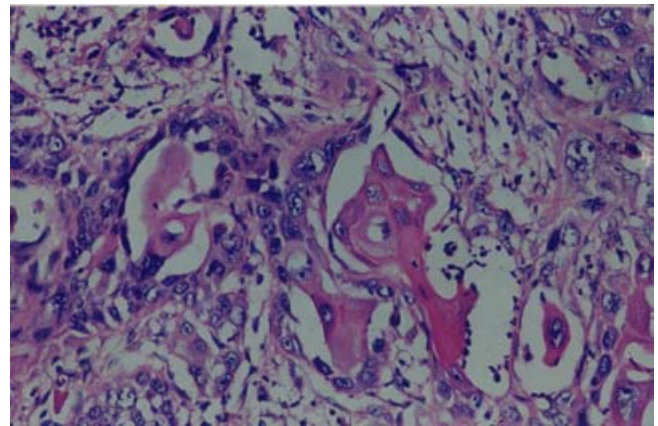


Figure 4
Photomicrograph of squamous cell carcinoma showing foci of keratinisation (Haematoxylin & Eosin \times 100)

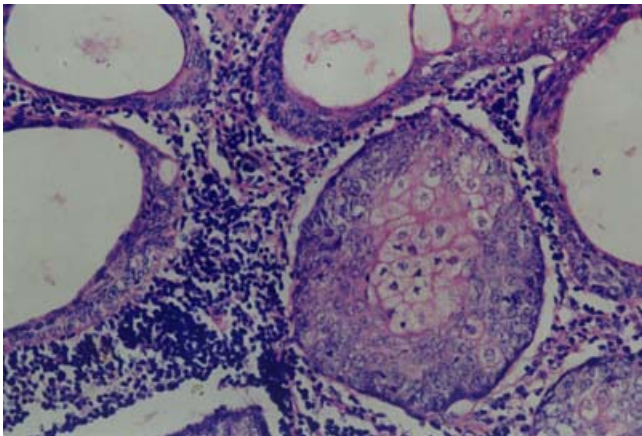


Figure 3
Photomicrograph showing duct like spaces lined by outer basaloid and inner mature sebaceous cells (Haematoxylin & Eosin \times 100)

On gross examination the parotidectomy specimen measured $7 \times 5 \times 2$ cm with irregular and nodular surface. Cut section showed grey white appearance with cystic spaces and foci of haemorrhage and calcification. Microscopic examination of the sections from salivary gland revealed partially circumscribed neoplasm (Figure 1) composed of numerous duct like structures and cystic spaces lined by flat to cuboidal epithelium and filled with keratin flakes intricately mixed with lymphoid stroma showing follicular formation in areas (Figure 2). Duct like space were

lined by outer basaloid cells and inner mature sebaceous cells were present (Figure 3) focal squamous and mucinous metaplasia of lining epithelium was also seen along with areas of fibrosis and calcification. In one area neoplasm was seen to arise from lining epithelium of duct like spaces composed of polygonal cells with moderate cytoplasm and hyperchromatic pleomorphic nuclei, arranged in sheets, nests and group foci of keratinisation seen in between cell groups (Figure 4). A diagnosis of sebaceous lymphadenoma with moderately differentiated squamous carcinoma was made.

Discussion

Sebaceous lymphadenoma is rare benign salivary gland neoplasm [1]. There have been several case reports describing the tumour and its association with other salivary gland neoplasm. Sebaceous lymphadenoma is predominantly seen in elderly females and age at presentation ranges between 25 to 89 years.

Microscopy in sebaceous lymphadenoma show variable sized sebaceous glands admixed with salivary ducts surrounded by dense lymphoid stroma. Lymphoid background has well developed germinal centres. Histiocytes, foreign body giant cell and inflammatory reaction are seen. Focal necrosis can be observed occasionally [2,3]

Histogenesis of Sebaceous lymphadenoma is unclear. Possible theories are that it develops within the ectopic salivary gland in intraparotid lymph node [1] or may arise due to sebaceous differentiation in other tumours [1,4]. Other tumours to consider with similar histological characteristic include tumours with focal sebaceous differentiation, Warthin's tumour, mucoepidermoid carcinoma,

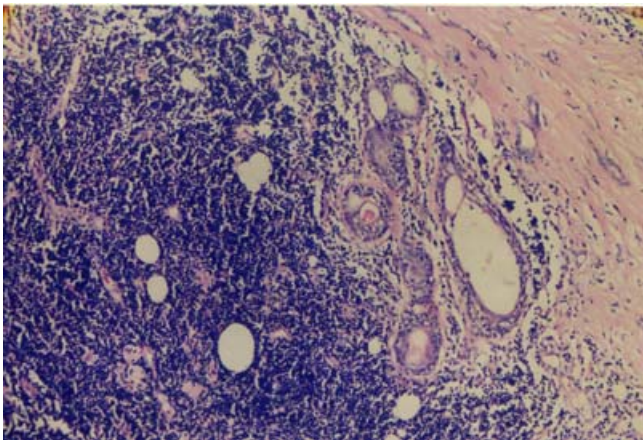


Figure 2
Photomicrograph showing neoplasm with intense lymphoid infiltrate (Haematoxylin & Eosin x 40).

pleomorphic adenoma (malignant), adenoid cystic carcinoma, benign oncocytoma and basal cell adenoma. [5,6]

Malignant transformation of the sebaceous lymphadenoma, although rare, should be considered along with possibility of a synchronous second primary malignant neoplasm in enlarging, locally invasive parotid lesions, considering that clinical behaviour and prognosis will be determined by the nature of the malignant component [7-9].

Parotidectomy is the treatment of choice in sebaceous lymphadenoma, however in presence of synchronous squamous cell carcinoma neck dissection should also be carried out if cervical lymph nodes are involved and we feel that postoperative adjuvant radiotherapy should improve the survival.

Conflict of interest

None declared.

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