

Sebaceous Adenocarcinoma of the parotid gland: a case report and literature review

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Citation

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Abstract

A 83 years old patient presented with a rapidly growing mass of the right parotid gland. Cytology was non-diagnostic, intra-operatively the tumor was found to invade the facial nerve and surrounding soft tissue structures. Resection of the parotid gland with facial nerve sacrifice was performed, yielding the final diagnosis of sebaceous adenocarcinoma. Sebaceous adenocarcinoma of the parotid gland is a rare and aggressive malignancy which can manifest as a solitary neoplasm in an otherwise normal gland, or appear concomitantly with other more common neoplasia such as pleomorphic adenoma or mucoepidermoid carcinoma. This entity and the relevant literature are reviewed.

CASE REPORT

An 83 year old male presented with a four months history of a right sided parotid gland mass. The mass had rapidly grown with no fever, or facial paresis, occasional night pain was noted. Past medical history included chronic renal failure, diabetes mellitus and mild CVA. Examination of the mouth and pharynx revealed medial bulging of the pharyngeal wall, without discharge from the Stensen's duct. The right parotid gland was enlarged, firm and non tender, without cervical lymphadenopathy. Computed tomography demonstrated diffuse enlargement of the right parotid gland, with involvement of the prestyloid parapharyngeal space. On fine needle aspiration cytology, epithelial and mesenchymal cells were observed. During the operation the tumor extended medial to the facial nerve & mandible, anteriorly the tumor involved the masseter muscle and reached the Stensen's duct. Due to the devastating extension of the tumor and involvement of the nerve until its peripheral branches, the facial nerve was sacrificed. Permanent section analysis revealed a lobular neoplasm composed of atypical tumor cells showing basaloid, squamous and sebaceous differentiation. Well formed ductal and glandular structures and perineural space invasion were identified (Figure 1 A, B), establishing the diagnosis of Sebaceous Adenocarcinoma. On 6 months follow up there was evidence of residual tumor infiltrated locally with additional new pulmonary lesion suspected for metastasis. Considering the aggressive progression of the disease, the patient's overall deteriorating status and his wish, further surgery or

radiotherapy were aborted, and he was referred to the oncology clinic. The patient died 4 months later due to aspiration pneumonia and sepsis.

Figure 1: Sebaceous carcinoma of parotid gland:

Figure 1

Figure 1A: Islands of sebaceous cells (arrow) surrounded by hyperchromatic cuboidal cells.

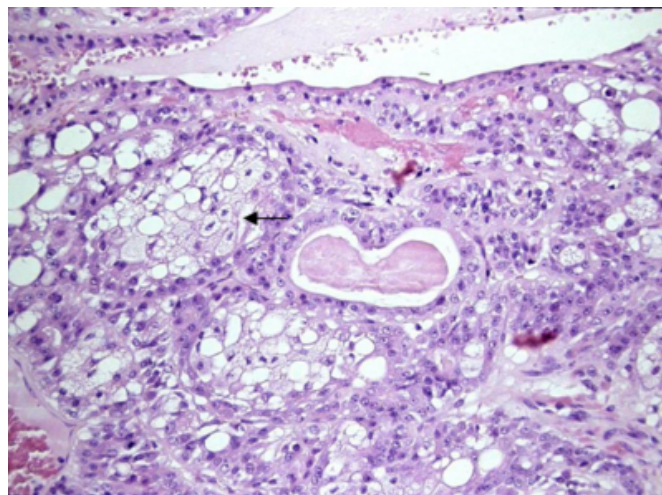
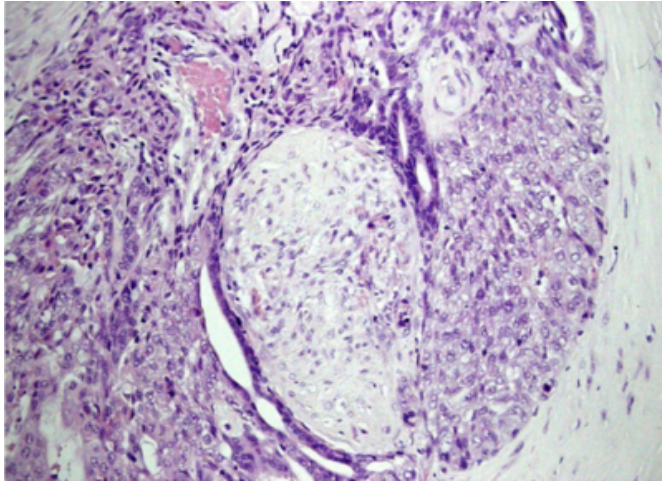


Figure 2

Figure 1B: Perineural space invasion.



DISCUSSION

Sebaceous adenocarcinoma is a rare malignancy mainly involves the ocular adnexae, skin and salivary glands. Up to date approximately 37 cases of salivary gland sebaceous carcinoma had been described, most of them involving the parotid gland and few appear in the submandibular and minor salivary gland¹²³. The age prevalence has two peaks, in adolescence and above the seventh decade of life, 2 cases were described in children⁴⁵. Clinically the symptoms vary from any indolent, slow-growing, painless and asymptomatic mass, to a painful rapidly progressing swelling accompanied by facial paralysis⁴⁵. If the sebaceous tumor is associated with lymphatic structures, it will be named lymphadenoma or lymphadenocarcinoma.

The etiology of sebaceous carcinoma in the salivary gland is in a longstanding debate. The first suggested mechanism is malignant transformation and sebaceous differentiation within a pleomorphic adenoma or mucoepidermoid carcinoma⁶⁷. Mesenchymal elements in benign mixed tumors of the salivary gland are thought to represent ectodermal products that have undergone metaplasia.

On the other hand, Linhartova⁴ and others postulated that sebaceous differentiation is a normal property of intercalated and striated salivary ducts. Therefore sebaceous neoplastic differentiation can be found in any type of salivary gland neoplasm which has ductal elements. Opinion holds that about one fourth of all surgically removed parotid glands, have some sebaceous elements. Nonetheless sebaceous glands are more numerous and better developed in parotid

glands where tumors had occurred¹⁸.

Another possibility is that sebaceous cells represent ectopic ectodermal structures not fully separated from the parotid gland. Ectopic sebaceous glands are frequently found in the head and neck areas, like Fordyce spots on the buccal mucosa and at mucocutaneous junctions³⁷. During embryogenesis there is an internal displacement of epidermal elements into the Parotid bud. The presence of accessory ectodermal structure such as the adenexae, may point to some ectopias in the salivary glands. Nevertheless, this theory does not explain the occurrence of sebaceous cells in the submandibular gland which is of endodermal origin².

The current hypothesis is that certain ducts in the salivary gland are pluripotential, which can differentiate into a variety of cells like sebaceous, squamous, oncocytic and mucous cells⁵. Therefore, the occurrence of sebaceous neoplasms in the salivary gland together with mucous or squamous cells, like was identified in our patient, does not necessarily point to the presence of another pre existing mixed or mucoepidermoid tumor. As expected clinical experience regarding this rare malignancy is limited, and different modalities of treatment are usually used. It seems reasonable to try and achieve local control of the tumor using wide local excision with or without neck dissection⁵⁷. Radiotherapy and chemotherapy have not proven to be effective in the treatment of sebaceous carcinoma⁷.

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