

Intraoral sebaceous carcinoma

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Abstract

Sebaceous carcinoma arising from the buccal mucosa is very rare. Only one such case has been reported in the English literature. We report a second case arising in an 11-year-old girl.

Key words: Sebaceous gland neoplasms; Mouth; Carcinoma, squamous cell

Case report

An 11-year-old girl had a lump excised from the left buccal mucosa at a local hospital nine months prior to the date she was seen in our hospital. A histology report was not available. The lump recurred six weeks after excision, and progressively increased in size. Biopsy was performed in another hospital and the patient was referred to our hospital with the diagnosis of carcinoma of the buccal mucosa. Other than a swelling in the left cheek and a painful ulcer in the left buccal mucosa, she had no other symptoms. On examination there was a 6 × 6 cm swelling in the left cheek, with the skin over the lesion. There was a 3 × 3 cm ulcerative lesion in the left buccal mucosa extending up to the upper gingivobuccal sulcus. Examination of the neck showed a 3 × 3 cm, hard, mobile, left submandibular lymph node. There was no trismus. The seventh nerve was intact. The rest of the ENT examination was normal. Biopsy was repeated and was reported as squamous cell carcinoma. Computer tomography (CT) scan confirmed that there was no involvement of the maxilla or the orbit. Metastatic workup did not show any evidence of lung, bone or liver metastasis. The patient underwent excision of the cheek tumour along with the overlying skin and radical neck dissection. The intraoral lining was reconstructed with a radial forearm flap and the defect in the cheek was reconstructed with a pectoralis major myocutaneous flap. Post-operative recovery was uneventful.

The operative specimen was reported as sebaceous carcinoma with excision margins clear (Figure 1). The left submandibular salivary gland was invaded by the tumour and two out of five lymph nodes in the submandibular region were positive for metastatic carcinoma. Post-operatively, the patient received a full course of radiation therapy to the primary and ipsilateral neck. There was no evidence of recurrence on her follow-up two years after surgery.

Discussion

Sebaceous glands are holocrine adnexal components of skin. They are widely distributed and in the head and neck

they are found in high density. They are normally found either closely associated with hair follicles (90 per cent) to form pilosebaceous apparatus, or independently in the skin.

Extracutaneous, extraocular sebaceous glands in the head and neck are principally found in the oral cavity and in major salivary glands (Batsakis and El-Naggar, 1990). Nearly 80 per cent of adults have clinically evident sebaceous glands in the oral mucosa (Halperin *et al.*, 1953; Miles, 1958).

Batsakis and El-Naggar (1990) classified sebaceous neoplasms of salivary glands as sebaceous lymphadenoma, sebaceous adenoma and sebaceous carcinoma. The two adenomas have a low recurrence potential, the carcinomas have a biological behaviour similar to that of ocular sebaceous carcinomas.

The sex distribution of sebaceous carcinoma has been reported to be variable. Urban and Winkelmann (1961), Beach and Severence (1942), and Miller and White (1967) described a higher incidence in men, but Rulon and Helwig (1974) found a predominance in white women. Warren and Warvi (1943) found a similar incidence in both sexes. This neoplasm most often occurs in older patients after the age of 40 years, and especially in the 60s and 70s. Our patient is probably the youngest to be presented.

Extraocular sebaceous carcinoma is an extremely rare tumour with fewer than 120 cases previously reported; of these approximately 75 per cent involved the head and neck. The most common site of origin was the parotid gland, representing 29 per cent of the total number of cases (Bailet *et al.*, 1992). Other sites of occurrence include face (Tchornobay *et al.*, 1992), nose (Motley *et al.*, 1991), external auditory canal (Doble *et al.*, 1981), retroauricular region (Guner *et al.*, 1991), hypopharynx (Martinez-Madrigal *et al.*, 1991) and buccal mucosa (Damm *et al.*, 1991).

Reports on intraoral sebaceous neoplasms are extremely rare in the English literature. Fewer than ten cases of intraoral sebaceous adenoma, and one case of intraoral sebaceous carcinoma have been reported (Bernier, 1959; Miller and McCrea, 1968; Epker and Henny, 1971; Lipani *et al.*, 1983; Gnepp and Brannon, 1984; Ferguson *et al.*, 1987; Orlian *et al.*, 1987; Damm *et al.*, 1991; Bailet *et al.*, 1992).

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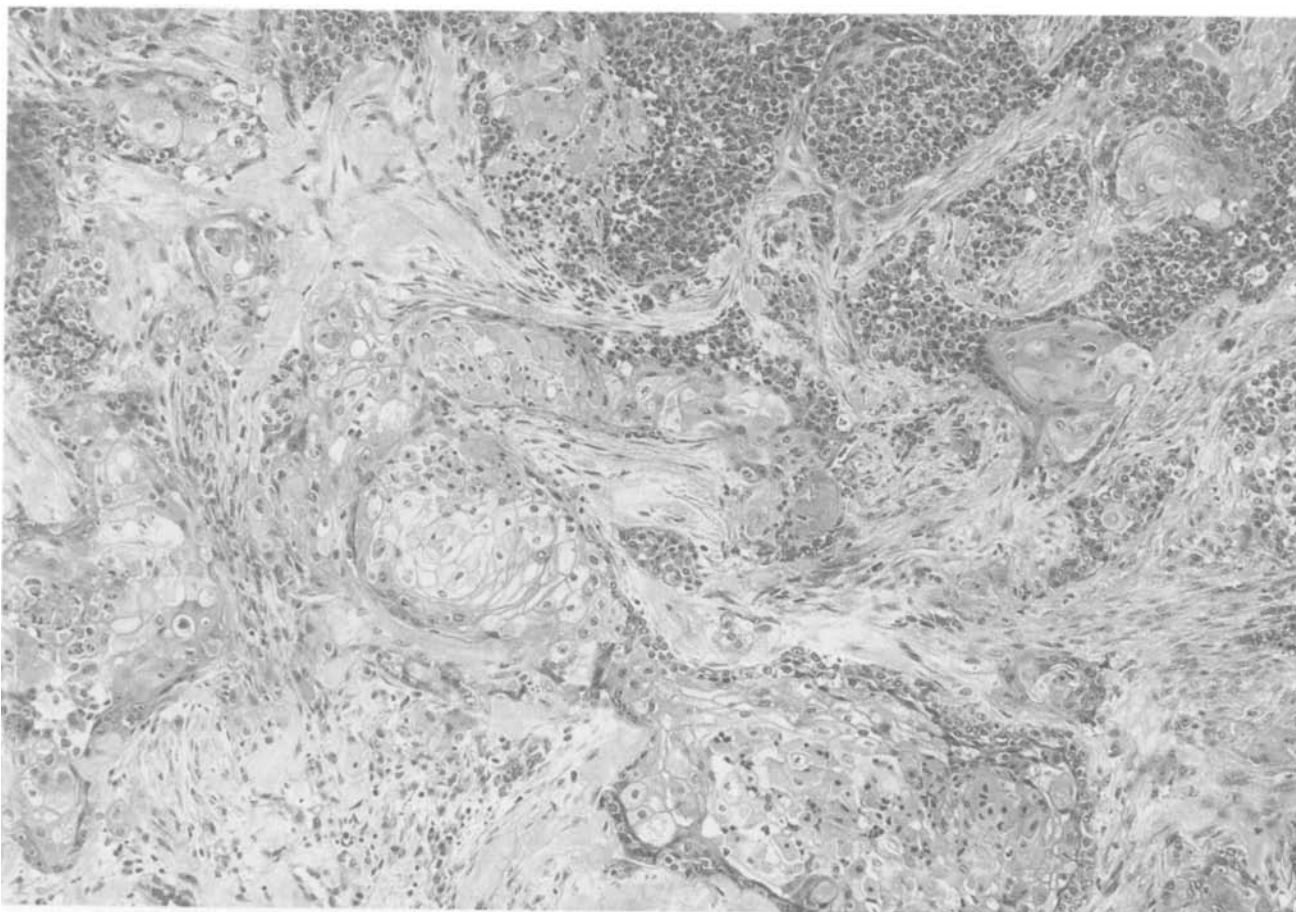


FIG. 1

Sebaceous carcinoma. Small cells with round nuclei are found in broad, solid areas (top) and lining the areas with sebaceous differentiation (bottom). The nuclei are round, uniform and dense. Nucleoli are not prominent. Cytoplasmic vacuolation is seen (H & E stain; $\times 80$).

Sebaceous carcinoma is usually a slowly growing tumour that is locally aggressive and capable of metastatic spread to regional lymph nodes and distant sites (Shulman *et al.*, 1973; King *et al.*, 1979; Wick *et al.*, 1985). Metastases to lymph nodes occurred in 21 per cent of patients in the series reviewed by Bailet *et al.*, 1992. Pulmonary, visceral and central nervous system metastases have also been documented (Rao *et al.*, 1982).

The characteristic histological features of sebaceous carcinoma include high mitotic activity, nuclear pleomorphism, lobular architecture and cytoplasmic vacuolization (Rulon and Helwig, 1974; Wick *et al.*, 1985). All neoplasms that contain clear-cells, such as poorly differentiated squamous cell carcinoma with hydropic swelling, clear-cell sweat gland carcinoma, and metastatic clear-cell renal carcinoma, should be differentiated from sebaceous carcinoma (Audisio *et al.*, 1987).

Wide surgical excision with frozen section monitoring of the margins to ensure adequate excision remains the main treatment modality for sebaceous carcinoma. A regional lymphadenectomy is warranted if the nodes are positive. Radiation therapy only with curative intent may be an effective treatment modality in early, less extensive lesions and may be used as an adjuvant in patients with metastases or for palliation (Bailet *et al.*, 1992).

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