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CASE REPORT

Oral squamous cell carcinoma in children; review of an unusual entity

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Summary Most commonly, squamous cell carcinoma (SCC) of the oral cavity presents during the fifth and sixth decades of life. Less than 4% of these cancers occur in patients younger than 40 years of age. Only a small sample of this subgroup exists of pediatric patients (≤20 years), making oral SCC in children an extremely rare entity. An 11-year-old boy is presented who developed a SCC of the gingiva. The relevant literature of oral SCC in pediatric patients will be reviewed as well.

1. Introduction

Squamous cell carcinoma (SCC) of the oral cavity is rare in patients of age 40 and younger, being primarily a disease that occurs in males in their sixth or seventh decade. Younger patients (aged less than 40 years) account for approximately 4% of all oral cancers [9]. Only a small sample of this subgroup exists of pediatric patients (≤20 years), making oral SCC in the pediatric age group an extremely rare entity.

The purpose of the present report is to describe an 11-year-old boy who developed a SCC of the gingiva, and to review the literature of oral SCC in pediatric patients.

2. Case report

An 11-year-old boy was referred to the Department of Oral and Maxillofacial Surgery by his orthodontist because of a non-tender, progressive growing swelling of the gingiva of the upper front teeth, noticed by the patient since 6 weeks. The relevant medical history did not reveal any abnormalities. The patient did not use any medication at presentation.

Physical examination revealed a firm, non-tender, verrucous swelling with indurated borders and central ulceration on the buccal and palatal aspects

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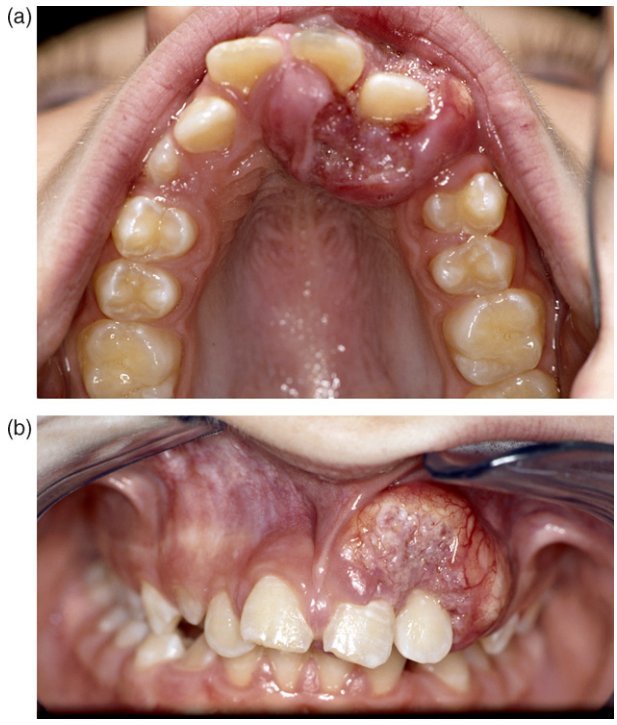

Fig. 1 A firm, non-tender, verrucous swelling with indurated borders and central ulceration on the buccal and palatal aspects of the left upper front teeth measuring 2.5 cm by 2.0 cm (a + b).

of the left upper front teeth measuring 2.5 cm by 2.0 cm (Fig. 1). The left upper incisors were mobile but tested positive on vitality tests. Multiple, non-tender, mobile submandibular lymph nodes were palpated bilaterally. On additional radiographic examination erosion, possibly infiltration of several millimeters of the alveolar bone between the two left upper incisors was observed. Histopathological examination of an incisional biopsy of the swelling showed a well-differentiated SCC. Because of the age of the patient there was some doubt about the diagnosis; a verruciform xanthoma with characteristics of pseudoepitheliomatous hyperplasia (PEH) was therefore considered as well (Fig. 2a).

A second biopsy specimen showed on microscopic examination again characteristics of a well-differentiated SCC (Fig. 2b). Based on fine needle aspiration cytology of the submandibular lymph nodes as well as a radiograph of the chest there were no signs of regional and/or distant metastasis. An additional computed tomogram revealed invasion of the buccal cortex of the left upper incisors (Fig. 3). Nineteen days after the initial presentation the tumor was diagnosed as a T4aN0M0 SCC of the gingiva of the left maxilla. The tumor was obviously grown in this short period (Fig. 4).

A partial maxillectomy was performed. The former diagnosis of a well-differentiated SCC was

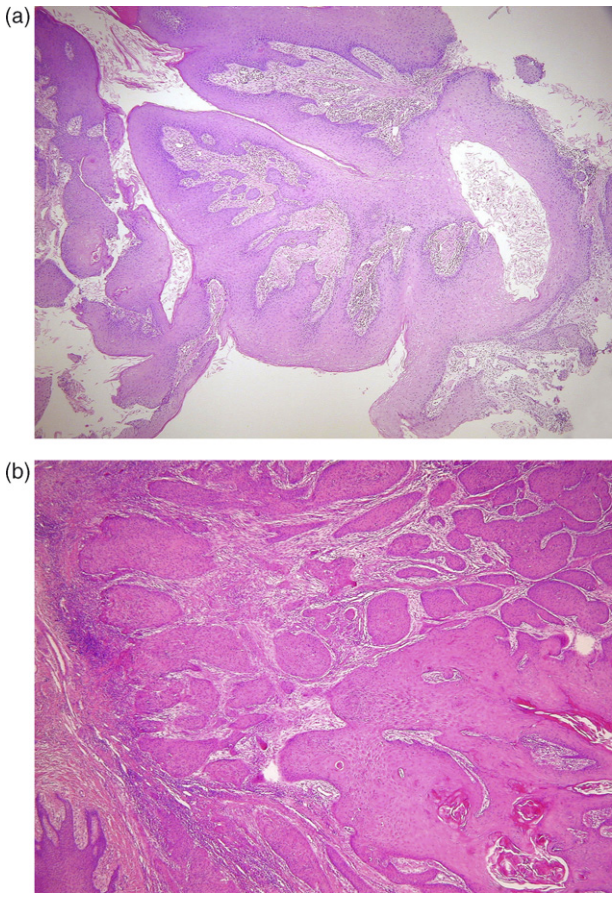

Fig. 2 (a) A biopsy specimen from the swelling shows histopathological features of a well differentiated SCC. A verruciform xanthoma with characteristics of pseudoepitheliomatous hyperplasia (PEH) was considered as well (haematoxylin—eosin, 10×). (b) A second biopsy specimen confirming the former diagnosis; a well differentiated SCC (haematoxylin—eosin, 10×).

confirmed on histopathological examination of the surgical specimen. The margins seemed to be free of tumor.

3. Discussion

Most commonly, SCC of the oral cavity presents during the fifth and sixth decades of life. Less than 4% of these cancers occur in patients younger than 40 years of age [9]. Only a small sample of this subgroup exists of pediatric patients (≤20 years), making oral SCC in children an extremely rare entity. The results of a literature review on oral SCC in pediatric patients from the period 1970—2005 have been summarized in Table 1 [1—5,7,8,10—18]. A total of 65 cases were found. There was no gender predilection. The majority of tumors occurred on the tongue, the lip or the gingiva. Patients' ages ranged from 2 to 20 years with only four cases being

Table 1 Summary of studies and case reports reporting oral squamous cell carcinoma in pediatric patients (≤20 years) (1970–2005)

Authors	Year	Country	No. of patients	Gender (M/F)	Age (years)	Site	Histology (differentiation)
Byers [3]	1975	USA	4	Unknown	17, 19, 19, 19	Tongue, *n* = 4	Well, *n* = 1; moderate, *n* = 1; poor, *n* = 1
Krolls and Hoffman [8]	1976	USA	19	Unknown	14, *n* = 3; 15–19, *n* = 16	Unknown	Unknown
Harper and Copeman [5]	1981	UK	1	1/0	18	Tongue	Poor
Yagi K et al. [18]	1981	Sudan	1	0/1	10	Tongue	Well
McGregor et al. [11]	1983	Canada	1	0/1	18	Tongue	Unknown
Newman et al. [12]	1983	USA	4	Unknown	14, 16, 18, 18	Tongue, *n* = 4	Unknown
Son and Kapp [15]	1985	USA	4	3/1	10, 17, 18, 19	Tongue, *n* = 2; cheek, *n* = 1; gingiva, *n* = 1	Well, *n* = 4
Sacks et al. [13]	1985	USA	1	1/0	13	Gingival	Well
Earle et al. [4]	1988	USA	1	1/0	7	Gingiva	Moderate
Keukens et al. [7]	1989	The Netherlands	1	1/0	9	Tongue	Well
Lund and Howard [10]	1990	UK	1	Unknown	20	Tongue	Unknown
Tsukuda et al. [17]	1993	Japan	4	2/2	14–19, *n* = 4	Unknown	Unknown
Sarkaria and Harari [14]	1994	USA	1	1/0	17	Tongue	Unknown
Atula et al. [1]	1996	Finland	1	1/0	19	Tongue	Unknown
Thompson et al. [16]	1999	USA	20	10/10	2–20	Tongue *n* = 9; lip *n* = 6; unknown *n* = 5	Well–poor
Bill et al. [2]	2001	USA	1	1/0	14	Gingiva	Well

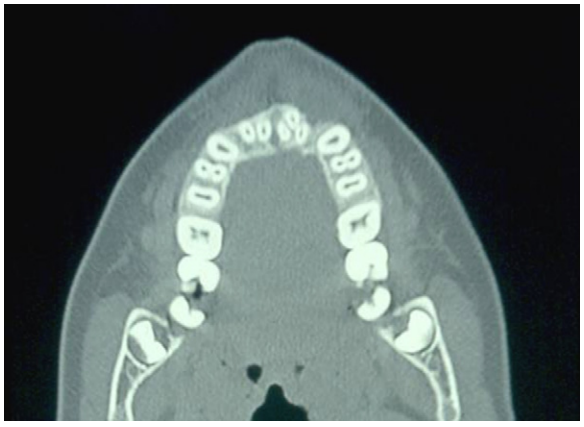

Fig. 3 A computed tomogram revealed invasion of the buccal cortex of the left upper incisors.

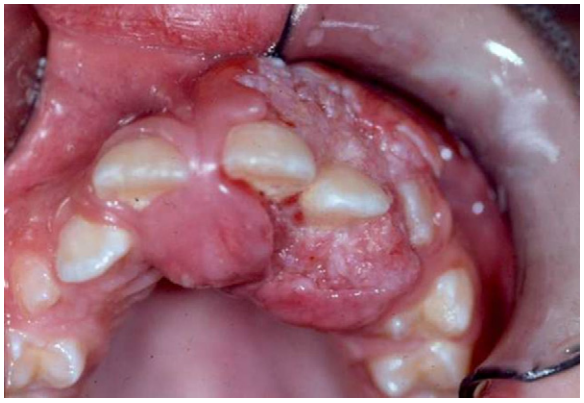

Fig. 4 Nineteen days after the initial presentation. The tumor was obviously grown in this short period.

younger than 12 years, making our case of an oral SCC in an 11-year-old boy unique.

Recently it has been observed that there is an increasing incidence of oral SCC in the younger population of several countries [6]. The role of traditional risk factors such as tobacco, alcohol, betel quid chewing, and low consumption of fruits and vegetables is unclear in this age group. Some studies have suggested that these patients may exhibit a predisposition to genetic instability.

There is a general trend in reported studies for SCC of the oral cavity in young patients to be particularly aggressive and carry a poorer prognosis than older patients [14]. A poorer prognosis in young patients could be due to (1) a differing disease etiology and tumor behavior or (2) delay in presentation and/or diagnosis (patients' and/or doctors' delay), the latter obviously playing a role in the currently presented case. The presentational and diagnostic delays may arise because of the reduced expectation of cancer in the young patient [6].

As inflammatory lesions in pediatric patients can become highly proliferative and assume neoplasm-like characteristics, the histopathologic diagnosis of SCC may present difficulties in this specific patient group. Pseudoepitheliomatous hyperplasia (PEH), a reactive process, may then be difficult to distinguish from SCC. Although differentiation between PEH and SCC in the adult patient group is generally not difficult for an experienced pathologist, PEH in pediatric patients may exhibit cellular atypia and irregular growth to such extend that this distinction may be less obvious. The difficulties that were encountered in our patient in attempting to histopathologically distinguish between PEH and SCC are comparable with the difficulties seen in several previously reported case of SCC in pediatric patients [4,13].

Although rare, SCC does occur in pediatric patients and should therefore be included in the differential diagnosis of inflammatory-like, rapid growing lesions of the oral cavity. PEH may be difficult to histopathologically distinguish from SCC in this specific patient group. Close collaboration between clinician and pathologist is therefore from utmost importance to achieve the assessment of a proper diagnosis.

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